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**Motor Competence and Quality of Life in Youth with Cancer and Visual
Impairments**

by

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For the Degree of Doctor of Philosophy in

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DEDICATION

It is with genuine gratitude that I would like to dedicate this dissertation to my family and friends. To my Mom, Dad, Maddy, and Anna for your unwavering support, distraction, and guidance. To Lizzie, for being my rock when I felt like I was drifting and for reminding me to go outside. Finally, to my Camp Abilities family, where each and every individual I have worked with at Camp has made an instrumental impact on me. I can only hope to one day reciprocate the love, support, and inspiration to you all.

With Love,

Emily

AKNOWLEDGEMENTS

Dr. Brian - Never did I think meeting a researcher at Camp Abilities would lead me to today. I have learned so much about myself as a teacher, researcher, and colleague and cannot thank you enough for presenting me with those opportunities. Thank you for providing me with the tools to navigate this process, the opportunity to grow, and for your unwavering belief in my potential. I look forward to what is yet to come.

Dr. Lieberman - Our journey began 10 years ago during my undergraduate degree at SUNY Brockport. Without your guidance, passion, and belief in not only individuals with visual impairments, but in me, I would not be where I am today. Thank you.

Dr. Stodden - Thank you for pushing me to think outside the box and to looking through a developmental perspective. I know that these skills will be of great value in my future.

Dr. Webster - With gratitude, I can confidently say that your way with words positively influenced my academic writing and know it will continue to do so long into the future.

Dr. Davis - Thank you for your medical expertise and opportunity to connect with, learn from, and research the youth with visual impairment sequela of cancer population.

Alex Stribing, Matthew Patey, Jenna Fisher, Adam Pennell, Sally Miedema-Taunton, and Hayes Bennett - I wish I had words to describe my thankfulness for each and every one of you. It is with your support, kindness, friendship, and camaraderie throughout these years that has led me to donning my Harry Potter garb today. I couldn't have done this without you, and thank you from the bottom of my heart.

ABSTRACT

This dissertation is comprised of a systematic review of the literature and two descriptive analytic studies, for a total of three studies. The overall purpose of this dissertation was to examine the motor competence of youth with pediatric cancers, a visual impairment (VI), and VI sequela of cancer (VISC), and to explore if motor competence and other variables in the physical domain predict overall health-related quality of life (HRQoL).

The purpose of Study 1 systematic review of literature was to explore and describe the motor competence of pediatric cancer patients and survivors in comparison to peers without cancer and to examine the correlates of motor competence for this population.

Methods: A full electronic search was completed on the following databases: MEDLINE, platform EBSCOhost, Web of Science, and Physical Education Index. The last search was completed March 25, 2018. The search limits were set from 2005 to present, language as English, scholarly peer-reviewed journals, and full-text. Inclusion criteria used to screen records were as follows: human participants, pediatric cancer diagnosis, participants between the ages of 1 to 18 years old at the time of cancer diagnosis; measure of gross motor competence; and intervention studies that included baseline motor data. No letters to the editor, reviews, or published abstracts were included. There was a total of 27 studies identified out of 2,180 that were initially screened. **Results:** The motor competence of pediatric cancer survivors has a tendency to be lower than their peers without a cancer diagnosis. The following correlates of motor competence were identified: level of physical

activity, body mass index and weight, grip strength, and age. Significant findings in this review were the gaps in the literature that were uncovered and included the following studies: perceived motor competence ($n = 0$), health-related quality of life ($n = 4$), and process-product measures ($n = 1$). To address these gaps in literature, future research should examine the differential effects and relationships on fundamental motor skills with process and product measures, perceived motor competence, and overall HRQoL in pediatric cancer survivors. **Limitations:** Further examination is necessary to generalize the results through larger sample sizes, valid, and reliable measures. **Conclusions:** The overall findings indicate low motor competence in pediatric cancer survivors compared to healthy peers and an association with level of physical activity, body mass index, grip strength, and age.

The purpose of Study 2 was to examine group differences between youth with a VI, VISC, and a healthy comparison group regarding HRQoL, motor competence, physical activity, and perceived motor competence. **Methods:** Participants ($N = 45$) had a mean age of 12.33 ± 2.65 years. Twenty-seven of the participants were male and 18 were female. There were three groups: VISC ($n = 15$), VI ($n = 15$), and a comparison group ($n = 15$). Each of the participants with a VISC was matched by age, degree of vision, and biological sex to participants in the other two groups. To examine group differences for HRQoL and perceived motor competence, a Kruskal Wallis Test with three groups (VI, VISC, and healthy peers) was used. Descriptive statistics, four (VI classifications) by three group (VISC, VI, and healthy peers) ANCOVAs, and Kruskal Wallis Tests with three groups were calculated to explore the mean differences. **Results:** The ANCOVAs revealed a significant difference between groups for TGMD-3 total scores ($F = 10.62, p < .001, \eta_p^2 =$

.34), product total combined z-scores ($F = 14.51, p > .001, \eta_p^2 = .41$), and physical activity ($F = 11.03, p < .001, \eta_p^2 = .35$). However, the Kruskal Wallis test revealed that the groups were not statistically different in HRQoL ($H = 2.10, p > .05$) or perceived motor competence ($H = 4.44, p > .05$). **Limitations:** The limitations of this study include a small sample size, a purposive convenience sample, possible testing site impact and potential biased responses due to the nature of self-reporting. **Conclusions:** The motor competence of youth with a VI and VISC have a tendency to be lower than their peers without a VI or VISC. Although, the motor competence of youth with VIs and a VISC were not significantly different from one another, future research should examine these differences further with a larger sample and examine the potential differences based on age at onset of VI.

The purpose of Study 3 was to examine the underlying mechanisms predicting physical activity and HRQoL for youth with a VI or VISC. **Methods:** For this study there were a total number of participants ($N = 30$) with a mean age of 12.33 ± 2.64 years and biological sex (male = 18, female = 12). Of the 30 participants there were two groups, VISC ($n = 15$) and VI ($n = 15$). Each of the participants with a VISC was matched by age, degree of vision, and biological sex. To examine the underlying mechanisms of physical activity and HRQoL descriptive statistics, Pearson's correlations, and two separate multi-level hierarchical regressions were used. **Results:** Physical activity was positively associated with product motor competence ($r = .71, p < .001$), TGMD-3 ($r = .60, p < .001$), HRQoL ($r = .42, p = .020$) and participants' degree of vision ($r = .58, p < .001$). HRQoL (PedsQLTM) was positively associated with TGMD-3 ($r = .37, p = .047$), product motor competence ($r = .53, p = .003$), perceived motor competence ($r = .42, p = .020$), physical

activity ($r = .42, p = .020$), biological sex ($r = -.37, p = .043$), and a co-morbidity ($r = -.42, p = .022$). Degree of vision, perceived motor competence, and motor competence accounted for 61 percent of the variance explained ($R^2 = .61, p = .005$) in participants' average step count scores, with motor competence accounting for 21 percent of the variance explained. A total of 51 percent of variance in participants' total HRQoL scores was explained by the presence of a co-morbidity, biological sex, perceived motor competence and motor competence, with motor competence adding an additional 16 percent of the variance explained ($R^2 \Delta = .16, p = .035$). However, physical activity did not significantly contribute to the variance explained in HRQoL. **Limitations:** The limitations of this study consisted of a small sample size, a purposive convenience sample, and possible testing site impact. **Conclusions:** The findings of this study indicate that the physical domain has a strong relationship with overall HRQoL for youth with a VI or VISC. Specifically, motor competence was the strongest predictor for both physical activity and HRQoL beyond participant characteristics.

The potential implications of this dissertation include the importance of motor competence in predicting youth's overall levels of physical activity and perceived HRQoL, which hold the possibility of having a large impact on health outcomes for youth with a VI and VISC. Based on the importance of motor competence, physical education has the potential to play an integral role in supporting the improvement and development of motor competence for youth with a VI or VISC.

Key Words: children, physical activity, fundamental motor skills, and perceived motor competence.

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CHAPTER 1

INTRODUCTION

Motor competence encompasses fine motor skills, balance, functional mobility, and fundamental motor skills (Robinson et al., 2015). Fundamental motor skills are the “building blocks” for more advanced movements including both locomotor skills (e.g., hopping) and ball skills (e.g., catching; Logan et al., 2018). Without a base knowledge of fundamental motor skills, an individual may be blocked by a proficiency barrier, preventing them from transferring their fundamental skills and motor competence to sport specific skills necessary for participating in physical activity throughout their lifetime (Clark & Metcalfe, 2002; Robinson et al., 2015; Seefeldt, 1980; True et al., 2017). Therefore, if children do not successfully learn their fundamental motor skills, they may be less likely to engage in physical activity opportunities throughout their lives (Robinson et al., 2015; Seefeldt, 1980; True et al., 2017).

Although motor competence is an important factor in estimated future physical activity participation, perceived motor competence may be a better predictor than any other construct beyond the presence of a disability (Brian et al., 2019b). Meaning, if a child does not believe they are proficient at a skill or task, they will be more likely to opt out of sport and lifelong physical activities. Therefore, motor competence and perceived motor competence are both important predictors to participation in physical activity, which

impacts body weight status (Babic et al., 2014; Brian et al., 2019b; Robinson et al., 2015; Stodden et al., 2008; True et al., 2017).

The physical domain that encompasses both motor competence and physical activity, as well as an individual's perceived motor competence, may predict long-term health-related quality of life (HRQoL). The World Health Organization (WHO) defines health as "a state of complete physical, mental, and social well-being, and not merely the absence of disease and infirmity" (WHO, no. 2, p. 100, 1948). HRQoL is a multi-dimensional construct of an individual's perception regarding their physical (e.g., playing at recess, riding on a bike), psychological (e.g., self-perception of body image and self-esteem), and social (e.g., playing with friends and family) well-being and functioning, which directs attention to the holistic impact of health on the personal well-being of an individual (Matza et al., 2004). The physical well-being dimension of HRQoL encompasses functional mobility, energy and vitality, physical function, physical activity, play (for children), and motor competence (Matza et al., 2004; Ravens-Sieberer et al., 2006). The psychological well-being dimension of HRQoL focuses on self-esteem, emotions, body image, cognitive ability, and perceptions (e.g., perceived motor competence; Matza et al., 2004; Ravens-Sieberer et al., 2006). The social well-being dimension of HRQoL encompasses social interactions, peer and parent relationships (Matza et al., 2004; Ravens-Sieberer et al., 2006). Based on the potential impact that motor competence, perceived motor competence, and physical activity may have on long-term HRQoL, it is imperative to examine these variables and their relationships with one another.

The tools used to measure motor competence, perceived motor competence, and physical activity are of utmost importance. Motor competence can be measured with tools that are process-oriented (e.g., Test of Gross Motor Development; TGMD), product-oriented (e.g., Bruininks-Oseretsky Test of Motor Proficiency [BOT-MP]), or a combination of the two. Process-oriented assessments measure the quality of the performance (e.g., rudimentary, functional, and mature) or components of the performance (e.g., steps in opposition; Burton & Miller, 1998). Product-oriented assessments evaluate distance, velocity, accuracy, height etc. for an outcome of the movement being performed (Burton & Miller, 1998). Researchers have recommended utilizing a combination of process- and product- oriented measures, as a more comprehensive assessment of motor competence to examine the variability and consistency of youth in different skill stages (Burton & Miller, 1998; Hands, 2002; Logan et al., 2016, 2018; Robinson et al., 2015; Rudd et al., 2015; True et al., 2017).

The ability to differentiate performance across a wide range of ability levels is essential when selecting a tool to assess youth with disabilities and/or diseases. For the purposes of this dissertation, motor competence, perceived motor competence, physical activity, and HRQoL are being explored in three populations of youth; pediatric cancer survivors, visual impairment (VI), and VI caused by cancer, as compared to their peers without a disability or disease.

Pediatric Cancer

Each day, approximately 43 children are diagnosed with cancer in the United States (American Cancer Society, 2018). Additionally, the rate of cancer diagnoses in the United States is increasing each year, along with an increase in five-year survival rates (American

Cancer Society, 2018). With the significant increase in survival rates, there is now a shift towards concerns for long-term HRQoL for survivors, and how their condition or treatment may impact their health over time (American Cancer Society, 2018). A diagnosis of cancer in childhood or adolescence abruptly disrupts daily life, including a reduction in the capacity for and involvement in recreational physical activity and sports (Götte et al., 2014; van Brussel et al., 2005). This interruption of daily life often leads to lower overall physical activity and total energy expenditure and is significantly correlated to unhealthy weight status in comparison to peers without a previous or current cancer diagnosis (van Brussel et al., 2005). The implications of low physical activity and unhealthy weight put pediatric cancer survivors at a greater risk of a poor HRQoL.

Overall, youth with cancer have significantly lower motor competence, physical activity levels, and are at a much greater risk of becoming overweight and obese than their peers without cancer (Augestad & Jiang, 2015; Florin et al., 2007; Green et al., 2013; Hartman et al., 2010; Hopkins et al., 1987; Leone et al., 2014; Ness et al., 2015; Piscione, et al., 2014; van Brussel et al., 2005; Weil et al., 2002). However, the vast majority of past research examining motor competence in youth with pediatric cancers utilized product-oriented measures, such as Movement Assessment Battery for Children (M-ABC) and BOT-MP, and only assessed participants with acute lymphoblastic leukemia (e.g., Hartman et al., 2008, 2010; Piscione et al., 2014). Perceived motor competence in the pediatric cancer population is also relatively unknown, and one study indicated that participants with acute lymphoblastic leukemia overestimated their actual motor competence (Hartman et al., 2008). For individuals with a previous diagnosis of cancer, physical activity, motor competence, balance, and muscular strength were positively associated with HRQoL (Ness

et al., 2015; Wright et al., 2005). The current literature is in need of substantially more evidence in regard to the physical domain, and particularly for participants with diagnoses other than acute lymphoblastic leukemia and the relationships with HRQoL.

Visual Impairments

In the United States, a VI is considered a low-incidence disability, and an estimated 656,389 school-aged youth diagnosed with a VI (CDCP, 2019). In regard to VIs, degree of vision (severity) is most commonly classified with visual acuity, visual field, and contrast sensitivity (American Printing House for the Blind, 2016; NASEM, 2016). For the purposes of this study degree of vision will be categorized based on an established sport-based classification system, B1 (e.g., no functional vision) to B4 (e.g., low vision; United States Association of Blind Athletes, 2017). A VI classified as B1 is defined as the lowest degree of vision and includes no light perception to some light perception without the ability to recognize the shape of a hand at any distance or in any direction. A classification of B2 includes the ability to recognize the shape of a hand with a visual acuity up to 20/600. The B3 classification includes a visual acuity of 20/600 – 20/200 and a visual field between 20 – 5 degrees. Finally, a classification of B4 is considered the highest degree of vision, and includes a visual acuity between 20/200 – 20/70 and a visual field larger than 20 degrees. All classifications are based on the best eye, including eye correction (e.g., glasses).

Youth with a VI often have lower motor competence, perceived motor competence, and physical activity participation, as well as higher rates of obesity, which appear to fit with Stodden and colleagues' model of the negative spiral of disengagement (Brian et al., 2016; Haegele et al., 2015; Houwen et al., 2007; Stodden et al., 2008; Wagner et al., 2013).

In contrast to the pediatric cancer literature, researchers examining the motor competence of youth with VIs have primarily utilized process-oriented measures, such as the Test of Gross Motor Development – 2 or – 3 (TGMD; e.g., Brian et al., 2016, 2018). We also know that there are many important factors positively associated with a high quality of life for individuals with VIs including independent travel, socialization, relaxation, sense of independence, and involvement in recreational physical activity (Ball & Nicolle, 2015; Columna et al., 2015). There is still a knowledge gap regarding the relationships between HRQoL with motor competence, and perceived motor competence.

Visual Impairment Sequela of Cancer (VISC)

Many pediatric cancer survivors – up to 22 percent – develop a permanent VI within ten years post treatment, that can be a result of the cancer itself or the treatment (Lackner et al., 2000). Of the youth diagnosed with the following cancers, the percent that may develop a long-term VI due to the tumor are: retinoblastoma (38%), brain tumors (24%), optic pathway glioma (22.5% to 69%; e.g., depending on tumor type and treatment; Campagna et al., 2010; De Blank et al., 2016; Desjardins et al., 2002; Macedoni-Lukšič et al., 2003). Furthermore, cancer treatments (e.g., external beam radiotherapy, and/or thermo- chemotherapy) may increase the likelihood of the development of a long-term VI in pediatric cancer survivors (Desjardins et al., 2002; Van Dijk et al., 2007). Brain tumors, which affect the central nervous system are the second most common type of cancer diagnosed in youth below the age of 20, and may cause a VI depending on the location of the tumor and/or the treatment (Packer et al., 2003). Retinoblastoma is another of the most commonly diagnosed childhood cancers and affects retinoblast cells in the retina, which can result in a VI (Desjardins et al., 2002). Optic Pathway Gliomas are a specific type of

infiltrative astrocytic tumor without defined borders that can interfere with the entire optic pathway, leading to varying degrees of vision loss and is most commonly diagnosed in children under 10 years of age (De Blank et al., 2016). Going forward, a VI that was caused by cancer or the treatment of cancer will be referred to as a VI sequela of cancer (VISC). A sequela can be defined as “an aftereffect of a disease, condition, or injury; or a secondary result” (Merriam-Webster Incorporated, 2019).

Moreover, a VISC (e.g., brain tumor, retinoblastoma) may further exacerbate the likelihood of negative effects on motor competence, physical activity, perceived motor competence, and the three dimensions of HRQoL as described in the pediatric cancer and VI populations above (Augestad & Jiang, 2015; Brian et al., 2016; Florin et al., 2007; Green et al., 2013; Haegele et al., 2015; Hartman et al., 2010; Hopkins et al., 1987; Houwen et al., 2007; Leone et al., 2014; Ness et al., 2015; Piscione et al., 2014; Stodden et al., 2008; van Brussel et al., 2005; Wagner et al., 2013; Weil et al., 2002). There are substantial gaps in the literature regarding the motor competence, perceived motor competence, and physical activity of individuals specifically with a VISC. Likewise, little is known about the HRQoL for youth with a VISC, though specifically for adults with an Optic Pathway Glioma (a VISC) HRQoL in the social and psychological domains are low and were not associated with age at onset of the glioma (De Blank et al., 2016).

In summary, youth with a VI or a previous diagnosis of cancer are more likely to have low motor competence, perceived motor competence, and physical activity putting them at risk for a poor HRQoL. The findings of lower motor competence, perceived motor competence, physical activity participation/rates and higher rates of obesity appear to fit with the Stodden and colleagues’ model of the negative spiral of disengagement for both

youth with a VI and youth with pediatric cancer (Brian et al., 2016; Florin et al., 2007; Haegele et al., 2015; Hartman et al., 2010; Hopkins et al. 1987; Houwen et al., 2007; Leone et al., 2014; Ness et al., 2015; Piscione et al., 2014; Stodden et al., 2008; van Brussel et al., 2005; Wagner et al., 2013; Weil et al., 2002). Despite the importance of the relationship between motor competence, perceived motor competence, and physical activity, much remains unknown about the associations among these variables for youth who have or had VI, or a VISC in comparison to their peers without either. Additionally, we do not know how motor competence, perceived motor competence, and physical activity associate with overall HRQoL in these populations of youth with a VI and a VISC.

For the purposes of this dissertation, we explored the differential effects of motor competence, perceived motor competence, physical activity, and HRQoL (physical well-being, social well-being, and psychological well-being) by group (VI, VISC, and a healthy comparison group). Additionally, we examined if the previous relationships conceptualized in the general population regarding the importance of motor competence and perceived motor competence in overall physical activity levels and body weight status still stands for these populations (Robinson et al., 2015; Stodden et al., 2008). Another extension of this dissertation was to explore if the variables in the 2008 model (Stodden et al., 2008) also predicts overall HRQoL for youth with a VI or VISC. Results from these studies will help to bridge the gap on what is currently unknown about youth with cancer, a VI, or VISC and address limitations of previous studies of youth with VIs or pediatric cancer survivors.

Purpose, Research Questions, and Hypotheses

Study 1 (systematic review of the literature). The purpose of this study was to conduct a systematic review of the literature exploring the gross motor competence of pediatric cancer patients and survivors in comparison to healthy peers, and the correlates of motor competence for this population (e.g., HRQoL, physical activity, perceived motor competence). The research questions for this study were as follows: (1) What is the motor competence of pediatric cancer survivors/patients, and (2) What are the correlates of motor competence for pediatric cancer survivors? Due to the exploratory nature of this study no hypothesis was made prior to investigation.

Study 2. The purposes of this study were to examine the differential effects of healthy peers, a VI, and a VISC on HRQoL, motor competence (process and product), perceived motor competence, and physical activity levels of youth. The research question for this study was: are there group differences between youth with a VI, a VISC, and healthy peers' groups on HRQoL, motor competence, physical activity, and perceived motor competence? The hypothesis for this study was that youth with a VISC, will have lower overall HRQoL, motor competence, perceived motor competence, and physical activity levels than their same age peers with only a VI or a healthy comparison group after controlling for co-morbidity.

Study 3. The purposes of this study were to examine the underlying mechanisms predicting physical activity in youth with a VI or a VISC, and to examine the underlying mechanisms predicting overall HRQoL in youth with a VI or a VISC. The research questions for this study were: (1) which of the factors (motor competence and perceived motor competence) predict physical activity in youth with a VI and VISC after controlling

for significantly correlated (to physical activity) participant demographics, (2) which of the factors (motor competence, perceived motor competence, and physical activity) predict body weight status in youth with VI and VISC after controlling for significantly correlated (to BMI) participant demographics, and (3) which of the factors (motor competence, perceived motor competence, and physical activity) predict HRQoL in youth with VI and VISC after controlling for significantly correlated (to HRQoL) participant demographics? The hypotheses for this study were: (1) motor competence and perceived motor competence will predict physical activity for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to physical activity), (2) motor competence, perceived motor competence and physical activity will predict body weight status for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to BMI), and (3) motor competence, perceived motor competence and physical activity will predict HRQoL for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to HRQoL).

Limitations and Delimitations

The limitations of this dissertation included the use of convenience sampling, site impact, self-report methodology, and potential lack of effort by participants on motor skills testing. The recruitment of the sample used was convenience sampling due to the feasibility of finding and working with a low incidence population. Therefore, the results of this study may not be generalizable. Additionally, with the population of youth with a VI or a VISC, a co-morbidity (e.g., autism spectrum disorder) may be present. Therefore, the presence of a comorbidity was controlled for in data analysis. While the sample size was a total of 45 participants for this dissertation, the only way this number could be reached was through a

large age range (e.g., 7-18 years), a variety of degrees of vision, and possible co-morbidities. In an attempt to address this limitation, participants with a VISC were all accepted then matched by age, biological sex, and degree of vision with youth with a VI and the comparison group (e.g., peers without a VI or VISC). Additionally, participant settings at a sports camp (VI and VISC groups) and a rural public school were a limitation of this study. When participants are completing self-report surveys they may respond with bias or in ways that reflect how they perceive the researchers may want or expect them to respond. To reduce the potential for this problem, the participants were asked to answer survey questions honestly in a private space. Effort and interest level in the movements can have a large impact on the study, some of the participants may not be excited about participating in some of the skills (e.g., underhand toss). To reduce potential effects of effort levels, the researcher worked to build an individual rapport with each participant, and encouraged the participants to do their best on each skill.

Significance, Innovation, and Rigor

This dissertation is significant because of the long-term potential to predict and improve the HRQoL for youth with a VI and a VISC through motor competence, perceived motor competence, and physical activity. This dissertation is innovative as it is the first to examine the motor competence, physical activity, perceived motor competence, and HRQoL of individuals with a VI and a VISC of cancer. This dissertation was conducted using rigorous methodology such as a combination of process and product measurements of motor competence as recommended by experts in the field.

The findings of this dissertation can lead to the long-term goal of improving HRQoL for individuals with a VISC in a multitude of ways. Initially by describing current

literature on gross motor competence of pediatric cancer patients and survivors in comparison to healthy peers, as well as the correlates of motor competence for this population (e.g., time since cessation of treatment). Followed by determining if group differences based on etiology for perceived motor competence, motor competence, HRQoL and physical activity are present and finally, by exploring the underlying mechanisms predicting physical activity and HRQoL for youth with a VI, a VISC, or without. With an understanding of the underlying mechanisms predicting physical activity and HRQoL for individuals with a VI and/or a VISC, it will be possible to design an intervention tailored to these populations to improve the mechanisms predicting physical activity and HRQoL.

CHAPTER 2:
STUDY 1 CORRELATES OF MOTOR COMPETENCE FOR PEDIATRIC
CANCER PATIENTS AND SURVIVORS: A SYSTEMATIC REVIEW

With the recent growth in survival rates of children diagnosed with cancer, the improvement in quality of life for child-adolescent survivors becomes increasingly important (American Cancer Society, 2018). HRQoL is a multidimensional construct containing physical, psychological, and social well-being and function (Matza et al., 2004; Ravens-Sieberer et al., 2006). Higher levels of physical activity have been found to be positively associated with HRQoL in the general population (Jalali-Farahani et al., 2020) and individuals with disabilities (Doja et al., 2018). In the general population, higher levels of motor competence and perceived motor competence demonstrate a positive relationship with physical activity levels (Robinson et al., 2015; Schwimmer et al., 2003; Stodden et al., 2008). Theoretically, youth with higher levels of motor competence are more likely to be physically active and fit throughout their lifespan, and therefore, more likely to avoid the negative health effects inadequate physical activity (Barnett et al., 2009; Clark & Metcalfe, 2002; Jaakkola et al., 2016; Lubans et al., 2010; Robinson et al., 2015; Stodden et al., 2008). Due to a higher incidence of obesity, low physical activity levels, and low motor competence for pediatric cancer patients/survivors (van Brussel et al., 2005), pediatric cancer patients/survivors are more likely to track into a negative spiral of disengagement in physical activity (Robinson et al., 2015; Schwimmer et al., 2003;

Stodden et al., 2008). Therefore, motor competence can indirectly affect HRQoL and warrants further exploration in youth with pediatric cancer.

Motor competence is an umbrella term encompassing fine motor skills, balance, functional mobility, and fundamental motor skills (Robinson et al., 2015). Throughout the literature fundamental motor skills have been considered the “building blocks” for more advanced or task specific movements including both locomotor skills (e.g., hopping) and ball skills (e.g., catching; Logan et al., 2018; Seefeldt, 1980). Suggesting that without a base knowledge of fundamental motor skills, an individual may encounter a proficiency barrier, further preventing them from sport or task specific skills throughout their lifetime (Clark & Metcalfe, 2002; Robinson et al., 2015; Seefeldt, 1980; True et al., 2017). Thus, if children do not successfully learn their fundamental motor skills, they may be at a greater risk of physical inactivity throughout their lives and indirectly impact their HRQoL.

Previous reviews on pediatric cancer patients or survivors have been conducted on the physical dimension of health. Example reviews include exercise interventions (Baumann et al., 2013), sports participation and physical activity (Götte et al., 2014), health-related fitness (van Brussell et al., 2005), balance (Turner et al., 2013; Varedi et al., 2017), and motor skill (Green et al., 2013). Three of the reviews focused on specific types of cancer, such as acute lymphoblastic leukemia (ALL) (Green et al., 2013; van Brussell et al., 2005; Varedi et al., 2017), leaving a gap in the literature for cancers not pertaining to ALL. Thus, the potential impact of motor competence on the whole multidimensional construct of HRQoL its relationships (e.g., time since treatment, physical activity) are relatively unknown for pediatric cancer patients’ and survivors without ALL. Therefore, this review will not only serve as an update to previous reviews on motor competence in

pediatric cancer survivors and patients but expand to different types of cancer beyond ALL and correlated of their level of motor competence.

A diagnosis of cancer in childhood or adolescents abruptly disrupts daily life including capacity for involvement in recreational physical activity and sports (Götte et al., 2014). From 1989 to 2009 there were seven studies that examined the gross motor competence of youth during or after treatment for ALL, generally reporting gross motor delays in the participants (Green et al., 2013). According to green and colleagues (2013) children with ALL demonstrated poorer gross and fine motor competence than healthy peers, with low levels of gross motor competence in 5% – 54% of the participants. However, we do not know if the findings from Green and Colleagues (2013) on motor competence levels with hold in youth with other types of pediatric cancer. Thus, the purpose of this systematic review was to examine the motor competence of all types of pediatric cancer patients and survivors in comparison to healthy peers and examine its association with other important variables (e.g., HRQoL, physical activity). For the purposes of this review, the definition of motor competence is adopted from Robinson and colleagues (2015): “‘Motor competence’ is a global term used in this paper to reflect various terminologies that have been used in previous literature (i.e., motor proficiency, motor performance, fundamental movement/motor skill, motor ability, and motor coordination) to describe goal-directed human movement” (p. 1274). The research aims for this review are:

1. Examine motor competence levels of pediatric cancer survivors/patients.
2. Examine associations among motor competence and other physical variables in pediatric cancer survivors.

Methodology

Search Strategy and Study Identification

This systematic review was completed following the guidelines of Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Moher et al., 2009). A full electronic search was completed on the following databases: MEDLINE, platform EBSCOhost, Web of Science, and Physical Education Index. The last search was completed March 25, 2018. The search limits were set to 2005 to 2018, and limited to full-text, peer-reviewed journals published in English. In addition to the electronic database search, forward (e.g., searching author) and backward (e.g., searching references of articles eligible) tracking was completed for all articles included in the review (Moher et al., 2009). Any additional articles that met the inclusion criteria, acquired through backward and forward tracking, were screened following the same procedures used for the initial database search. The search strategy included three lines of search terms separated by ‘AND’, within each line all terms were separated by ‘OR’. See Table 2.1 for all terms included in the search strategy. Three of the researchers reached consensus on the complete search strategy before the initial search began.

Table 2.1. Search Strategy Terms for all Databases

| |
|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| motor skill, motor performance, motor competence, gross motor, fundamental motor skills, motor development, motor fitness, motor ability, motor coordination, fundamental movement, motor function, athletic skill, athletic competence, motor proficiency, object control, balance, locomotor skill, object manipulation, motor fitness |
| cancer, oncology, leukemia, tumor, malignancy, sarcoma, melanoma, lymphoma, myeloma, tumor, leukemia, Hodgkin’s disease |
| children, pediatrics, child, youth, juvenile, toddler, adolescent, teen, youngster, minor, young people, kid. |

Study Selection

Inclusion and Exclusion Criteria. The inclusion criteria used to screen articles were as follows: human participants; participants between the ages 1 to 18 years old with a pediatric cancer diagnosis; a measure of gross motor competence; intervention studies with baseline motor competence data were included. Exclusion criteria used to screen articles were as follows: activities for daily living or adaptive functioning; studies that only assessed fine or visual motor competence; outside of age limits (1-18 years), studies that used surveys or questionnaires as their primary measurement of motor competence. Additionally, letters to the editor, reviews and published abstracts were not included in the review.

Study Screening. The Initial screening process was completed by one researcher, where duplicates were removed and titles were screened for inclusion and exclusion criteria, then reviewed and agreed on by a second researcher. Throughout all remaining stages of the screening process, identified articles were reviewed independently by two researchers, and any disagreement was resolved with a third researcher. With the initial database search, backward, and forward tracking 2,180 studies went through the initial screening, 2,012 were included for further screening after removing duplicates. The screening process included reviewing the title, abstract, and then the full text of the remaining records and excluding records that did not meet the inclusion or exclusion criteria.

Study Eligibility. Next, all titles were screened to eliminate studies that clearly did not meet eligibility (i.e., not using human participants or different disabilities). Of the 103 studies remaining after the initial title screen, abstract screening eliminated an additional

71 studies, leaving 32 to be screened by full text. An additional five articles were removed for the following reasons: ($n = 2$) no gross motor scores provided, ($n = 1$) a protocol paper, ($n = 1$) a review of the literature, and ($n = 1$) participants did not have a cancer diagnosis (Figure 2.1; Moher et al., 2009).

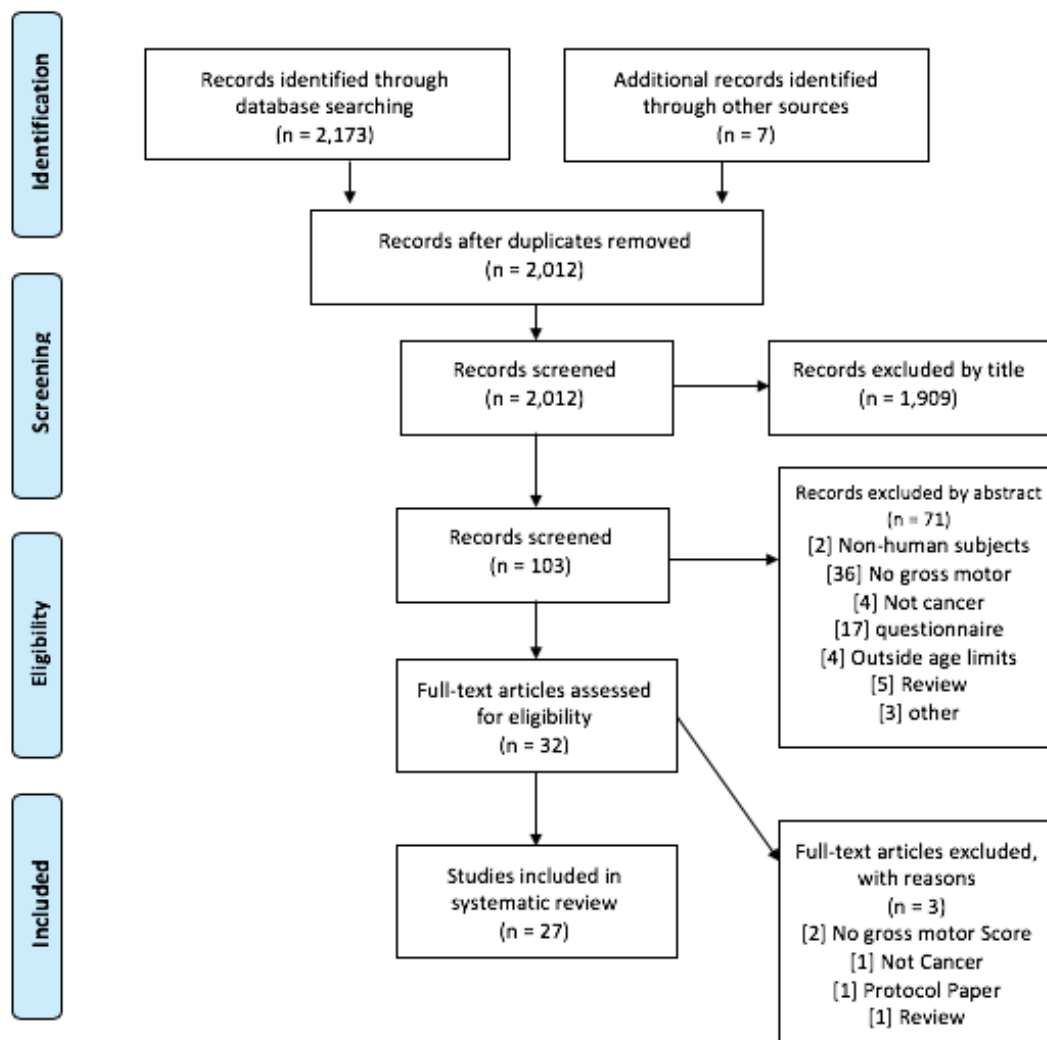


Figure 2.1. Flow Diagram for this systematic search

Data Collection Extraction and Analysis

The data extraction process was completed independently by one researcher then independently checked by a second researcher; any disagreement was resolved by a third researcher. To do this we developed a data extraction excel spread sheet, with one sheet

for risk of bias and one sheet for data directly related to the research questions. The following information was extracted as present (coded as a 1) or not present (coded as a 0) for risk of bias in the included studies: clear description of study purpose, clear description of study design, inclusion & exclusion criteria, detailed description of participants, description of the setting, control or comparison group, detailed description of assessments used, described validity & reliability of measures, detailed description of data analysis, provided explicit information for replication, limitations clearly stated, and statistical findings clearly stated. The following data was extracted directly related to the research questions: number of participants, participants diagnosis, participants age, participants biological sex, participants age at diagnosis, participants BMI or height and weight, participants time since treatment, study setting, when the study took place during or post treatment, did they longitudinally track the participants (coded, 0 = no, 1 = yes), sample total gross motor competence scores, and sample subscale motor competence scores. Then to answer the second question correlations or differences in motor competence based on the following were also extracted: treatment, fitness scores, physical activity levels, and HRQoL. The results extracted from the included studies were then qualitatively synthesized and descriptively reported with study frequency and percent.

Results

Study Characteristics

Risk of Bias. All of the studies ($N = 27$) provided information on the motor competence assessment and procedures; however, in many studies' validity ($n = 9$) and reliability ($n = 12$) data for assessments used was not explicitly documented. Only four studies included ethnicity data, only four were blinded to group coding, and none

provided socioeconomic status of the participants. The complete risk of bias evaluation of the included studies can be viewed in Table 2.2.

Table 2.2. Risk of Bias Evaluation Criteria for Included Studies.

| | Frequency (N = 27) | % |
|-----------------------------------------------|---------------------------|----------|
| Clear description of study purpose | 26 | 96.3 |
| Clear description of study design | 24 | 88.9 |
| Inclusion & exclusion criteria | 27 | 100 |
| Detailed description of participants | 26 | 96.3 |
| Description of the setting | 21 | 77.8 |
| Control or comparison group | 12 | 44.4 |
| Detailed description of assessments used | 27 | 100 |
| Described validity & reliability of measures | 14 | 51.9 |
| Detailed description of data analysis | 25 | 92.6 |
| Provided explicit information for replication | 25 | 92.6 |
| Limitations clearly stated | 21 | 77.8 |
| Statistical findings clearly stated | 24 | 88.9 |

Participants and Settings. Of the 27 articles included in the review, seven were conducted in the US, six in the Netherlands, five each in Germany and Canada, two in Australia and one each in Sweden and Malaysia. The total number of participants diagnosed with cancer included in this review was 1,145. To the best of our knowledge ($n = 1,111$) are independent of one another, with one study consisting of a longitudinal follow up (Hartman et al., 2013). Motor competence testing took place in the included studies during ($n = 8$ studies), and post treatment ($n = 18$), with one study having participants both during and post treatment (Beulertz et al., 2013). Of the studies that took place post cessation of treatment a majority took place within zero to five years ($n = 12$). However, two studies examined participants five to ten years, four studies had larger ranges of time since treatment (1 – 17 years), and one study did not disclose how long since cessation of treatment (Beulertz et al., 2013). Multiple cancer types were noted across studies including: ALL ($n = 826$), Wilms Tumor ($n = 74$), Brain Tumor ($n = 74$), Non-Hodgkins Lymphoma

($n = 38$), malignant mesenchymal tumor ($n = 26$), Bone Tumor ($n = 21$), Lymphoma ($n = 10$), Other Solid Tumor ($n = 10$), Osteosarcoma ($n = 9$), Central Nervous System ($n = 9$), Ewing Sarcoma ($n = 8$), Germ Cell tumor ($n = 6$), Acute Myeloid leukemia ($n = 5$), bone marrow transplant ($n = 4$), Rhabdomyosarcoma ($n = 3$), Nephroblastoma ($n = 2$), Rare disease ($n = 2$), Neuroblastoma ($n = 1$), Skin Tumor ($n = 1$), Tissue Sarcoma ($n = 1$), Sarcoma ($n = 1$), Spinal Cord Glioma ($n = 1$) and not specified ($n = 13$). The break down within each study can be seen in Table 2.3.

Motor Competence Assessments. The overall frequency of motor competence assessments in the 27 included studies were as follows: Movement Assessment Battery for Children (M-ABC; $n = 7$ studies), Bruininks-Oseretsky Test of Motor Proficiency Second Edition Short Form (BOT-2 SF; $n = 5$ studies), Bruininks-Oseretsky Test of Motor Proficiency Second Edition (BOT-2; $n = 4$ studies), Bruininks-Oseretsky Test of Motor Proficiency Second Edition – Balance Subscale (BOT-2 Balance; $n = 3$ studies), The Motor-Proficiency-Test for children between 4 and 6 years (MOT 4 – 6; $n = 3$ studies), German Motor-Test 6-18 (DMT 6-18; $n = 3$ studies), Gross Motor Function Measure (GMFM; $n = 2$ studies), MOON Test ($n = 2$ studies), and other ($n = 6$ studies). Multiple of the studies utilized more than one motor competence assessment ($n = 9$ studies). When examining the motor competence assessment tools used 26 of the studies used product-oriented tools and the remaining study, used a process-oriented tool (Naumann et al., 2015).

Major Findings

Motor Competence in Youth with Pediatric Cancer. Motor competence delays or significantly lower scores than norms or a comparison group were found in the in the following domains: hand-eye coordination (Götte et al., 2015; Van Brussel et al., 2006),

Table 2.3. Study Participant Demographics and Instrumentation

| Study | N | Diagnosis | Age (years) | Height /Weight (BMI) | Sex (M) | Motor Assessment |
|---------------------------|----|---------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------|-----------------------------|---------|----------------------|
| (Beulertz et al., 2013) | 26 | 11 NHL, solid tumor 6, lymphoma 5, CNS 2, Rare disease 2 | 9.09 | | 17(9) | MOT 4-6, DMT 6-18 |
| (Beulertz et al., 2016a) | 13 | 4 ALL, 2 Lymphoma, CNS 3, other tumor 4 | 11.14 (3.53) | | 8(5) | MOT 4-6, DMT 6-18 |
| (Beulertz et al., 2016b) | 33 | ALL 10, Germ Cell tumor 6, Rhabdomyosarcoma 3, CNS 4, NHL 2, Hodgkin Lymphoma 2, Nephroblastoma 2, Neuroblastoma 1, Skin Tumor 1, Tissue Sarcoma 1, Ewing Sarcoma 1 | 8.7(4.67) | 133.95cm/35.13 kg | 17(16) | MOT 4-6, DMT 6-18 |
| (De Luca et al., 2013) | 37 | ALL | 2.5-5* | | 21(16) | M-ACB 2, BOT- 2 SF |
| (Esbenshade et al., 2014) | 17 | ALL | 5-10* | | 2(10) | BOT-2 SF |
| (Gohar et al., 2011) | 9 | ALL | Med 4.0* | | 6(3) | GMFM |
| (Götte et al., 2015) | 47 | AML 5, ALL 13, Ewing Sarcoma 7, Osteosarcoma 9 | 12.6(3.9) | 19% overweight * | 20(27) | MOON Test |
| (Hartman et al., 2013) | 31 | ALL | 12.3 | 12% obese, 35% overweight * | 15(19) | MABC- 1&2 |
| (Hartman et al., 2006) | 12 | ALL 57, WT 43, NHL 12, MMT 8 16 | 8.1(2.2) | | 50(78) | M-ABC |
| (Hartman et al., 2008) | 92 | ALL 44, WT 28, NHL 10, MMT 10 | 8.9 | | 32(60) | M-ABC |
| (Hartman et al., 2010) | 34 | ALL | 7.8(2.2) | | 16(18) | M-ABC |
| (Hartman et al., 2009) | 51 | ALL | Med 5.3 & 6.2* | | 21(30) | Bayley scales, M-ABC |
| (Hooke et al., 2016) | 13 | Not Specified | 10 -13 | | 10(3) | BOT- 2 Balance |

| | | | | | | |
|-----------------------------|----|---------------------------------------------------------------------------------|------------|-----------------------------|--------|-----------------------------------------------------|
| (Hung et al., 2017) | 13 | ALL | 9.6(1.4) | 19.2(3.6) | 4(9) | BOT-2 SF |
| Kesting et al., 2015) | 21 | Bone Tumor | 15.2(2.1) | | 8(13) | MOON Test |
| (Leone et al., 2014) | 20 | ALL | 10.56(.65) | | 10(10) | UQAC-UQAM test Battery |
| (Marchese et al., 2017) | 5 | ALL | 6 – 17* | | 6(4) | BOT-2 Balance, Vertical Jump Task |
| (Naumann et al., 2015) | 26 | 17 ALL, BMT 4, WT 3, Brain Tumor 3, Lymphoma 1, Sarcoma 1, Spinal Cord Glioma 1 | Med 6.91* | | 15(11) | Department of Education and Get Skilled: Get Active |
| (Ness et al., 2015) | 10 | ALL | Med 10 * | Mean percentile 57.6 (3.15) | 38(71) | BOT-2 SF |
| (Piscione et al., 2014) | 30 | Brain Tumors | 11.4 | 8% overweight, 8% obese | 15(15) | BOT- 2 |
| (Piscione et al., 2017) | 28 | Brain Tumor | 11.53 | 25% overweight, 14%obese | 12(16) | BOT-2 |
| (Sabel et al., 2016) | 13 | Brain Tumor | 12.5(2.9) | | 7(6) | BOT-2 |
| (Tanner et al., 2017) | 13 | ALL | 5.79 | | 79(56) | Peabody Developmental Motor Scales 2, BOT 1& 2 |
| | 5 | | | | | |
| (Tay et al., 2017) | 10 | ALL | 4.8-18.0* | | 34(67) | BOT-2 BF |
| | 1 | | | | | |
| (van Brussel et al., 2006) | 13 | 12 ALL, 1 T-NHL | 15.5(5.8) | (20.75) | 7(6) | M-ABC |
| (Wiernikowski et al., 2005) | 10 | ALL 8, NHL 2 | Med 7.2 * | | 1 (9) | BOT-MP, GMFM |
| (Wright et al., 2005) | 99 | ALL | 12.1(4.9) | 141.9(17.7)/43.2 (18.8) | 44(55) | BOT-MP Balance |

Note: * = No means or *SD* were provided. Diagnosis acronym: ALL = Acute Lymphoblastic Leukemia, WT = Wilms Tumor, Brain Tumor (BT) ($n = 74$), NHL = Non-Hodgkins Lymphoma, MMT = Malignant Mesenchymal Tumor, CNS = Central Nervous System, AML = Acute Myeloid leukemia, BMT = Bone Marrow Transplant

control (Beulertz et al., 2013), speed (Beulertz et al., 2016a, 2013; Götte et al., 2015; Sabel et al., 2016), agility (Piscione et al., 2014; Tanner et al., 2017), body coordination (Beulertz et al., 2016a; Sabel et al., 2016; Piscione et al., 2014; Tanner et al., 2017), balance (Beulertz et al., 2016a; Götte et al., 2015; Marchese et al., 2017; Piscione et al., 2014; Sabel et al., 2016; Wright et al., 2005), and ball skills (Hartman et al., 2008; van Brussel et al., 2006). Although there were some inconsistencies in data reported across studies, the most commonly found delays were in hand-eye coordination, ball skills, balance, body coordination, speed, and agility.

The overall findings of the included studies consistently reported that a diagnosis of pediatric cancer is often followed with overall low levels of motor competence in comparison to normative data and healthy comparison groups (Beulertz et al., 2013; Beulertz et al., 2016a; Götte et al., 2015; Hartman et al., 2006, 2008, 2009; Naumann et al., 2015; Tanner et al., 2017; van Brussel et al., 2006; Wright et al., 2005). According to De Luca et al. (2013) and Hartman et al. (2010) 26% - 29% of the sample were delayed in their motor competence, in contrast Leone and colleagues (2014), found almost 50% of the participants were below the 15th percentile. The variance in results between the participants motor competence delays in the studies could be attributed to the measurement tools selected, or the participants age differences at testing (in each of the three studies the participants had ALL; De Luca et al., 2013; Hartman et al., 2010; Leone et al., 2014).

Motor Competence by Assessment. Overall, using the M-ABC 1 and/or 2, 25% - 54% of the participants scored below the 15th percentile (De Luca et al., 2013; Hartman et al., 2010, 2008, 2006, 2013; van Brussel et al., 2006). Mean scores for children with pediatric cancer on the BOT-2 Short Form were between the 23rd-53rd percentile, and 1%-

16% were below the 15th percentile (Esbenshade et al., 2014; Tay et al., 2017; De Luca et al., 2013; Hung et al., 2017; Ness et al., 2015). For the studies that used the BOT- 1 or - 2 Balance, all studies reported between 66% - 70% of the participants scoring below average mean scores for children with pediatric cancer (Wright et al., 2005; Marchese et al., 2017; Hooke et al., 2016). Additionally, for the entire BOT-2, below average z-scores were reported for children with pediatric cancer (25% sample with z-scores = -1 to -2), except fine manual control in one study (Piscione et al., 2014, 2017; Sabel et al., 2016). Out of three studies that used the MOT 4 – 6 and DMT 6 – 18, two found significantly lower motor competence scores compared to normative scores, one study did not (Beulertz et al., 2016a, 2016b, 2013). A few assessments were less commonly used (e.g., Peabody Developmental Motor Scales 2, Bayley scales). Results from these assessments indicated the motor competence of pediatric cancer survivors was significantly lower (up to 16% below the 15th percentile and 25% sample with z-scores = -1 to -2) than the normative scores (Esbenshade et al., 2014; Tay et al., 2017; De Luca et al., 2013; Hung et al., 2017; Ness et al., 2015; Piscione et al., 2014, 2017; Sabel et al., 2016).

Relationship Between Motor Competence, Cancer Diagnosis, and Treatment

Differences regarding levels of motor competence may vary based upon cancer type (Hartman et al., 2006; Kesting et al., 2015; van Brussel et al., 2006). The most common cancer type examined in the studies was ALL ($n = 826$), making up 72% of the participants across the studies. In two studies, children diagnosed with ALL were found to have no significant difference in motor competence in comparison to other cancer diagnoses ($p = .599$; Beulertz et al., 2013), Götte et al. (2015) found bone tumor patients to have lower

mean scores in motor competence than ALL patients although not significant (only 16 participants completed coordination of upper extremity assessment).

In addition to the cancer itself studies examined the relationships and differences in motor competence by the patient's treatment. The only treatment reported as significantly correlated to motor competence was cranial irradiation (Adjusted $R^2 = .187$, $p = .001$; Wright et al., 2005). However, the following cancer treatments did not significantly differ in their motor competence scores: treatment for ALL on participants with polymorphisms of CYP3A5, MDR-1 or the vincristine toxicity related MAPT gene did not differ significantly in their motor competence scores (Hartman et al., 2010), vincristine or no vincristine ($p = .494$; Beulertz et al., 2013), and duration of chemotherapy treatment at least two years post treatment (Tay et. al., 2017).

Correlates of time since treatment and participant age with motor competence

Time since the cessation of treatment was often not correlated to motor competence. Overall, a majority of the studies ($n = 4$ studies out of $n = 7$ studies) that examined time since treatment and motor competence did not report a significant relationship or difference (Hartman et al., 2006; Leone et al., 2014; Piscione et al., 2014; van Brussel et al., 2006). Two studies reported that participants were demonstrating a significant decrease in the levels of motor competence the longer since treatment (De Luca et al., 2013; Kesting et al., 2015). One study reported five years post intervention, motor competence significantly improved in ALL patients (Hartman et al., 2013). Up to six years post treatment participants still were demonstrating low levels of motor competence (Hartman et al., 2006; Leone et al., 2014; Piscione et al., 2014; van Brussel et al., 2006), suggesting permanent damage or a need to intervene. The findings across the included studies varied in the

relationship or differences of age to/in motor competence, one study reported age differences in motor competence in comparison to normative scores (not significant at 0 to 6 years old, but significant 6 – 17 years, $p = .000$; Beulertz et al., 2013) and one study reported age to be correlated to motor competence ($r = .54, p = .034$; Götte et al., 2015). In contrast three studies reported no association between age and motor competence (De Luca et al., 2013, Hartman et al., 2006; Leone et al., 2014).

Correlates among HRQoL, Physical Activity, Fitness, and Motor Competence

Physical activity, HRQoL, body mass index, and fitness have been correlated to motor competence for pediatric cancer survivors (Götte et al., 2015; Ness et al., 2015; Piscione et al., 2017; Sabel et al., 2016; Wright et al., 2005). Specifically, physical inactivity during treatment was correlated with low motor competence (Götte et al., 2015; Piscione et al., 2017), but one study found that motor competence was not correlated with physical activity (Hung et al., 2017). Additionally, body weight and body mass index were associated with motor competence (Götte et al., 2015; Piscione et al., 2017), and balance scores (Götte et al., 2015; Wright et al., 2005). Fitness components were also reported to be associated with motor competence, specifically low scores in grip strength and wrist strength adversely affected hand function and ball skills (Hartman et al., 2008). Motor competence for individuals with a previous diagnosis of cancer had a positive association with HRQoL in three studies (Gohar et al., 2011; Ness et al., 2015; Wright et al., 2005). However, Beulertz and colleagues (2013) found no significant differences between pediatric cancer participants and a healthy comparison group in HRQoL.

Discussion

The empirical evidence in this systematic review indicates that, overall, the motor competence in pediatric cancer survivors is lower than that of same-aged peers without a cancer diagnosis. Although a majority of the participants (72%) in the 27 studies were youth with ALL, 14 studies examined the motor competence of youth with other types of cancer (see Table 2). Within this review, potential correlates of motor competence for pediatric cancer survivors include physical activity, body mass index, weight, grip strength, and age (Götte et al., 2015; Ness et al., 2015; Piscione et al., 2017; Sabel et al., 2016; Wright et al., 2005). However, further examination is needed (e.g., larger sample sizes, differential cancer diagnoses, longitudinal, and experimental designs) in order to more effectively examine correlates. Gaining a more in-depth understanding of the correlations to motor competence for youth with pediatric cancer is essential for the development of effective interventions in the long-term for these children. Possibly the most important finding of this review is that time since cessation of treatment was either not correlated with, or negatively correlated with, motor competence, meaning children with pediatric cancer were often below the norm regardless of the amount of time since treatment ended (De Luca et al., 2013, Hartman et al., 2006; Kesting et al., 2015; Leone et al., 2014; Piscione et al., 2014; Van Brussel et al., 2006).

Limitations of the studies reviewed include a lack of generalizability, due to small sample sizes and a majority of the studies still exclusively examining participants with ALL ($n = 14$). As noted in the risk of bias evaluation, studies reported limited information about validity and reliability of measurements used, and two of the interventions did not provide enough information to be replicable. Caution is recommended in interpreting the

results of this review, given that a quantitative meta-analysis was not conducted. Additionally, only studies that written in the English language were included in the review, leaving a possibility of some studies in other languages not being included.

Physical educators can potentially play a key role in future trajectories of HRQoL through indirectly developing motor competence for this vulnerable population. In a study by Hartman and colleagues (2006), 91% of the participants received Physical Education and 65% of the participants still scored below the 50th percentile of norm scores in motor competence. Therefore, physical education teachers should complete motor assessments for all students that are pediatric cancer survivors returning to school. Then tailor their teaching towards the individual needs of their students based on a the motor competence assessment conducted, particularly referring the student for adapted physical education services if they score below the 15th percentile. Based on the results of this review, physical education teachers should tailor their programs to enhance the most common lower motor competence subscales (hand-eye coordination, balance, body coordination, ball skills, speed, and agility) for pediatric cancer patients and survivors (Beulertz et al., 2016a, 2013; Götte et al., 2015; Hartman et al., 2008; Marchese et al., 2017; Piscione et al., 2014; Sabel et al., 2016; Tanner et al., 2017; van Brussel et al., 2006; Wright et al., 2005).

This review of the literature exposed many gaps in research yet to be filled. Future research in this area needs to adequately document relevant information on tools used, measurement validity and reliability, and participants' demographics (e.g., race/ethnicity and socioeconomic background) to enhance the context of the findings. Specifically, children diagnosed with a VISC were excluded from all studies in this review, leaving unknown the impact a VISC may have on their motor competence and should be explored

in the future. Only one study in the review utilized a process-oriented assessment to examine motor competence, additional studies should employ process measures or both process and product tools to examine participants' motor competence holistically. Furthermore, future interventions are needed to focus on improving motor competence of youth with pediatric cancer, as a potential mechanism for improving overall HRQoL as indicated by the relationships found in the included studies (Gohar et al., 2011; Ness et al., 2015; Wright et al., 2005). In these interventions, physical activity and social interaction should be implemented through fun, developmentally appropriate motor tasks (e.g., fundamental, sport, or recreation specific motor tasks) (Beulertz et al., 2013; Hartman et al., 2006; Götte et al., 2015). Research implementing an experimental design need to clearly address descriptions of both intervention and control conditions and treatment fidelity.

In conclusion, the motor competence of pediatric cancer survivors has a tendency to be lower than peers without a cancer diagnosis. Differences regarding levels of motor competence may vary based upon cancer type (Hartman et al., 2006; Kesting et al., 2015; van Brussel et al., 2006), assessment used (De Luca et al., 2013), and time since treatment (De Luca et al., 2013). Additionally, the positive association between motor competence and HRQoL in pediatric cancer survivors should continue to be examined as a possible mechanism for improve the participants' long-term quality of life.

CHAPTER 3:
STUDY 2 DIFFERENCES IN MOTOR COMPETENCE, PHYSICAL ACTIVITY,
PERCEIVED MOTOR COMPETENCE AND HRQOL FOR YOUTH WITH A
VISUAL IMPAIRMENT WITH AND WITHOUT CANCER

The World Health Organization defines HRQoL as an “Individuals’ perception on their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards, and concerns” (WHO, 2011). The multidimensional construct of HRQoL encompasses physical well-being and function, psychological well-being and function, and social well-being and function (Matza et al., 2004; Ravens-Sieberer et al., 2006). Multiple benefits of physical activity have been demonstrated in all three domains of HRQoL; for example, physical (e.g., body weight status), psychological (e.g., enjoyment), and social (e.g., developing friendships; CDCP, 2020). Within the general population for children and adolescents, motor competence and perceived motor competence are consistent predictors of physical activity (Robinson et al., 2015; Stodden et al., 2008). Although there is a developing knowledgebase of HRQoL, physical activity, motor competence, and perceived motor competence in the general population, limited evidence exists to indicate how these variables may differ for youth with a VI, particularly youth with a VI as a result of a cancer\ . It is important to explore the physical dimension (physical activity, motor competence) and its correlates (perceived

motor competence) in youth with a VISC, because of the overall impact the physical domain may have on long-term HRQoL.

According to Lackner and Colleagues (2000), 22% of pediatric cancer survivors develop a VI sequela of cancer (VISC). A sequela is defined as “an aftereffect of a disease, condition, or injury; or a secondary result” (Merriam-Webster Incorporated, 2019). The likelihood of developing a long-term VI can be even higher in cancers that directly impact the eye (Ocular VI), brain and/or central nervous system (Cerebral VI) such as retinoblastoma (38%), optic pathway glioma (22.5% to 69%; e.g., pending on tumor type and treatment), and brain tumors (24%; Campagna et al., 2010; De Blank et al., 2016; Desjardins et al., 2002; Macedoni-Lukšič et al., 2003). Treatments received to combat cancer can also cause a VISC (e.g., chemotherapy; Whelan et al., 2010).

Youth with VI consistently show delays in motor competence, perceived motor competence, and physical inactivity (Brian et al., 2016; Haegele et al., 2015; Haegele & Porretta, 2015; Houwen et al., 2007; Wagner et al., 2013). Often, pediatric cancer survivors also have delays in both motor competence and perceived motor competence (Green et al., 2013; Lackner et al., 2000). However, these delays were found among youth with acute lymphoblastic leukemia and therefore cannot be generalized to youth with other types of cancers, leaving a large gap in the current literature. Additionally, pediatric cancer survivors and youth with VIs are at greater risk for poor HRQoL compared to their same aged healthy peers (Boulton et al., 2006; Chadha & Subramanian, 2011; Gohar et al., 2011; Habib & Irshad, 2018; Ness et al., 2015; Wong et al., 2009; Wright et al., 2005).

Not only is generalizability a concern regarding youth with cancer, there is also a lack of information incorporating a combination of process- and product- oriented

measurements encompassing youth with both VIs and pediatric cancers. While process-oriented assessments evaluate the quality of the performance or components of the performance, product-oriented assessments measure the outcome of the movement being performed (Burton & Miller, 1998). Utilizing a combination of product-and process-oriented measurements allows for the differentiation of variability in performers of different skill levels, creating a more comprehensive assessment (Burton & Miller, 1998; Hands, 2002; True et al., 2017). Utilizing this combination of process- and product-oriented measures of motor competence has been a recent recommendation by many experts in the field (Logan et al., 2016, 2018; Robinson et al., 2015; Rudd et al., 2015; True et al., 2017).

There is a knowledge gap regarding motor competence in youth with pediatric cancers other than acute lymphoblastic leukemia (Gilbert et. al., In Preparation). Knowledge regarding the motor competence of youth pediatric cancer survivors that did not have acute lymphoblastic leukemia is needed, because we cannot assume that the implications will be the same. Additionally, it is important to note that all participants with a VISC have been excluded from studies examining motor competence of youth pediatric cancer survivors. This leaves a substantial amount of the population's motor competence implications unknown. This is alarming due to the fact that many types of cancer and the treatment itself may lead to a VISC in up to 22% of pediatric survivors (Lackner et. al., 2000).

Furthermore, studies investigating youth with VIs have not identified or separated participants by etiology, such as a VISC. A diagnosed VISC may further exacerbate the likelihood of negative effects on motor competence, perceived motor competence, PA

levels, and HRQoL. However, this remains unknown in the literature. Therefore, the purpose of this study was to examine group differences between youth with a VI, VISC, and a healthy comparison group regarding HRQoL, motor competence, physical activity, and perceived motor competence.

Research Question 1. Are there group differences between youth with VI, VISC, and a healthy peers comparison group on HRQoL, motor competence, physical activity, and perceived motor competence beyond the presence of a co-morbidity?

Hypothesis1. Children with a VISC will have the lowest HRQoL, motor competence, perceived motor competence, and physical activity levels, followed by children exclusively diagnosed with a VI after controlling for co-morbidity.

Methodology

Participants and Setting

A purposive convenience sample of youth (7-18 years old) was recruited from summer camps for children who are blind and visually impaired. These camps were located in the following states: New York, Delaware, Texas, and Florida. Due to the low incidence of this population a convenience sample was necessary for the feasibility of this study. The comparison group was recruited from a rural low socioeconomic school district in New York, December 2019.

A total of 45 participants were included in this study in three groups, VISC ($n = 15$), VI ($n = 15$), and comparison group ($n = 15$) with a mean age of 12.33 ± 2.65 years, biological sex (male = 27, female = 18), race/ethnicity (86.4% Caucasian, 4.5% Asian, 4.5% Black, 4.5% other), and body mass index ($M_{BMI} = 21.30 \pm 5.47$). Some of the participants in this study also had a co-morbidity ($n = 10$). The specific diagnoses of the

co-morbidity ($n = 1$ participant had two) were as follows: other health impairment ($n = 3$), hearing impairment ($n = 2$), traumatic brain injury ($n = 2$), emotional disturbance ($n = 1$), developmental delay ($n = 1$), autism spectrum disorder ($n = 1$), and multiple disabilities ($n = 1$). The majority of the participants VIs were congenital ($n = 17$), with the rest being acquired VIs ($n = 13$). See Table 3.1 for participant demographics by group.

Table 3.1. Descriptive Statistics for Demographic Variables by Group

| | VISC | | VI | | Comparison | |
|------------------|-------------|------|-------------|------|-------------|------|
| | M(SD) | | M(SD) | | M(SD) | |
| Age (years) | 12.47(2.85) | | 12.20(2.51) | | 12.33(2.74) | |
| Body mass index | 22.67(6.07) | | 20.08(5.33) | | 21.11(4.94) | |
| | <i>N</i> | % | <i>N</i> | % | <i>N</i> | % |
| Biological Sex | 9(Male) | 60.0 | 9(Male) | 60.0 | 9(Male) | 60.0 |
| Co-Morbidity | 6 | 40.0 | 4 | 26.7 | - | - |
| Congenital VI | 5 | 33.3 | 12 | 80.0 | - | - |
| Degree of Vision | | | | | | |
| B1 | 7 | 46.7 | 8 | 53.3 | - | - |
| B2 | 4 | 26.7 | 3 | 20.0 | - | - |
| B3 | 2 | 13.3 | 2 | 13.3 | - | - |
| B4 | 2 | 13.3 | 2 | 13.3 | - | - |

All participants with VISC were included in this study, and participants from the VI and comparison groups were matched for age, biological sex, and degree of vision. Degree of vision (severity) is most commonly classified with visual acuity, visual field, and contrast sensitivity (American Printing House for the Blind, 2016; NASEM, 2016). For the purposes of this study, degree of vision will be categorized based on an established sport-based classification system, B1 (e.g., no functional vision) to B4 (e.g., low vision), in this system all classifications are based on the best eye including eye correction (USABA, 2017). Specifically, B1 classification is the lowest degree of vision (e.g., includes no light perception to light perception), B2 classification (e.g., visual acuity up to 20/600), B3 classification (visual acuity between 20/600–20/200 and a visual field between

20 – 5 degrees), and B4 classification is considered the highest degree of vision (e.g., visual acuity between 20/200 – 20/70 and a visual field greater than 20 degrees; USABA, 2017).

Instrumentation

Health-related quality of life. HRQoL was measured using the Pediatric Quality of Life Inventory™ Version 4.0 Short Form 15 (PedsQL™ 4.0 SF15) survey (Varni et al., 1998). Three different age bands of the PedsQL™ 4.0 SF15 – SF were used including: Teen Report 13 – 18, Child Report 8 – 12, and Young Child Report 5 – 7. Each of the age bands are comprised of 15 items and four dimensions. The four dimensions of the survey include physical functioning (5 items), emotional functioning (4 items), social functioning (3-item), and school functioning (3 items; Varni et al., 1998). The participants answered each prompt on a five-option Likert scale, 0 (never) to four (almost always; Chen et al., 2005). Surveys are scored by reverse scoring and transforming each item onto a scale of zero to 100 (e.g., 0 = 100, 4 = 0), then dividing by the number of items in the dimension; a higher score indicates a superior perception of HRQoL (Varni et al., 1998, 1999). The PedsQL™ 4.0 SF15 – SF Generic Core Scale results have demonstrated acceptable validity and reliability for pediatric HRQoL with the following internal consistencies using Cronbach's Alpha: total score ($\alpha = .82$), physical health ($\alpha = .60$), emotional functioning ($\alpha = .76$), social functioning ($\alpha = .74$), and school functioning ($\alpha = .75$; Chen et al., 2005).

Motor competence. The Test of Gross Motor Development third edition (TGMD-3) was used to measure process motor competence including locomotor and ball skills subscales (Webster & Ulrich, 2017). The locomotor subscales include the following skills: run, hop, skip, gallop, jump, and slide (Webster & Ulrich, 2017). The ball skills subscale includes: two-handed strike, one-handed strike, one-handed dribble, two-handed catch,

kick, overhand throw, and underhand throw (Webster & Ulrich, 2017). All scored trials are coded with either a one (met criteria) or zero (did not meet the criteria). The validity and reliability psychometrics using the TGMD-3 are strong for youth with VIs, up to 19 years of age (Brian et al., 2018). Modifications specifically for individuals with VIs were made for implementing the TGMD-3, such as utilizing a beep kickball and a beep baseball (Brian et al., 2018). According to Webster and Ulrich (2017) the TGMD-3 can be used as a criterion reference for all ages, assuming an individual does not earn a perfect score.

Product scores were collected on the following TGMD-3 motor skills: hop (distance), jump (distance), kick (velocity), and throw (velocity; Bürgi et al., 2011; True et al., 2017; Stodden et al., 2014). To measure the velocity product outcomes, maximum miles per hour for throw and kick was recorded using a Stalker radar gun (Stalker Radar, Plano, TX; Stodden et al., 2014). Participants were asked to hop from cone to cone (5 meters apart), for two scored trials. Three consecutive hops total distance (heel – to – heel) in centimeters are to then be recorded as the product outcome for each trial using video analysis software (Dartfish Video Analysis Software, Version 7.0). Standing long jump was measured for two trials from the takeoff line to the back of the closest heel on landing, which was recorded onsite with a tape measure to the nearest centimeter (Stodden et al., 2014). Then the average distance of the two standing long jump trials was used for analysis.

Perceived motor competence. The Test of Perceived Motor Competence for children with VIs (TPMC-VI; ages 7 to 8; Brian et al., 2016), Self-Perception Profile for Children (ages 9 to 13 years; Harter, 2012b), or the Self-Perception Profile for Adolescents (ages 14 to 19 years; Harter, 2012a) were used in this study to measure perceived motor competence in all participants (Brian et al., 2016, 2018). The TPMC-VI is a modified

version of the Pictorial Scale of Perceived Competence and Social Acceptance, utilizing vignettes to describe the content in the photos for youth with VI (Brian et al., 2016; Harter & Pike, 1984). The TPMC-VI is a six-item questionnaire with a two-question, forced-choice response, based on a specific motor skill situation (e.g., “really true for me” or “sort of true for me”). The scoring for this measure is a scale from one to four, with one being low and four being high. The Self-Perception Profile for Children and the Self-Perception Profile for Adolescents follow the same format as the TPMC-VI except that there are only five items on the adolescent scale. All three measures should require between five to ten minutes to complete. The psychometric properties of TPMC-VI reveal strong content and face validity and reliability for individuals with a VI (Brian et al., 2016). Both of the batteries (Self-Perception Profile for Children and the Self-Perception Profile for Adolescents) have moderate to strong psychometric properties and consistently produce results that are considered valid and reliable (Harter, 2012a, 2012b).

Physical activity. Physical activity data were collected using Fitbit Inspire™ and Fitbit Ace2™ activity trackers. The Fitbit Inspire™ was used with children 13 years of age and older and the Fitbit Ace2™ was used with children 12 years of age and younger. The Fitbit Inspire™ and Fitbit Ace2™ wearable activity trackers have a sensor that uses a tri-axial accelerometer to measure steps taken, distance traveled, calories burned, active minutes, hourly activity and stationary time (Fitbit™, San Francisco, CA). Additionally, the Fitbit Inspire™ and Fitbit Ace2™ devices are capable of submersion in water up to 50 meters, allowing participants to wear them during aquatic activities. The Fitbit™ devices stored seven days’ worth of minute-by-minute detailed motion data that was downloaded for analysis (Fitbit™, San Francisco, CA).

Procedures

University Institutional Review Board (IRB) approval and approval from external entities (camps and school districts) was obtained prior to data collection. Throughout the study all IRB rules, regulations, and training requirements were adhered to. Upon arrival to camp, parents and participants completed parental consent and child assent before being enrolled in the study. All assessments for individuals with a VI and VISC were administered in the months of June, July, and August 2019. Then all assessments for the comparison group were administered in January of 2020. The assessments were conducted in a gymnasium, open room, or a designated outdoor space at the summer camps or in the school district. The assessments were administered in the same order at each camp and school district to ensure assessment order was not a factor in the results. However, the testing sites for the different groups may have been a factor in the findings of this study, particularly with the educational sports camp not being a typical week of physical activity for the participants in the VI and VISC groups. However, due to the low incidence of this population a convenience sample located at the sports camp was necessary to get a large enough sample size for this study.

Participants were assigned and provided a Fitbit Inspire™ or Fitbit Ace2™ to collect their physical activity data throughout the week. Participants were instructed to wear the Fitbit Inspire™ or Fitbit Ace2™ on their non-dominant hand for four to six days, for a minimum of ten hours per day (Pate et al., 2004; Puyau et al., 2002). The span of days the activity tracker was worn depended on whether data were collected from a participant at a summer camp, and the duration of that camp, or during one week of school.

Participants' height and weight were measured while parents completed the demographic questionnaire. Participants' actual standing height and weight were measured using a portable stadiometer and digital scale. Demographic information was collected using a questionnaire, completed by the participants parent/guardian, that included participants' birth date, height, weight, race/ethnicity, biological sex, degree of vision, self-reported VI diagnosis, self-reported cancer diagnosis, and age at onset of VI and/or cancer diagnosis.

A measurement of perceived motor competence (TPMC-VI, Self-Perception Profile for Children, or Self-Perception Profile for Adolescents) and HRQoL (PedsQLTM 4.0 SF15) was then completed. Each participant was provided the appropriate age-specific assessment for perceived motor competence and HRQoL. The participants were asked to answer the prompts, either one-on-one with a trained coach or independently (large print and braille copies were provided). Each survey took between five and fifteen minutes to complete.

The final assessments administered were the motor competence measurements (TGMD-3 and product scores). All motor competence measurements were video recorded for coding purposes. Interrater reliability was calculated prior to data coding using an intraclass correlation coefficient for ball skills subscale ($ICC = .968$, 95% $CI = .94 - .99$, $p < .001$) and locomotor subscale ($ICC = .949$, 95% $CI = .90 - .98$, $p < .001$). For each gross motor skill, the equipment, preparation and protocol from the TGMD-3 (Webster & Ulrich, 2017) were used, except for modifications for VI used from Brian and colleagues (2018). For each skill, demonstration and/or verbal instructions were provided. The product data were collected on the same trials following the TGMD-3 protocol. Participants were

instructed to perform their best (e.g., “Jump as far as you can”) to facilitate effortful performance for each motor skill (Langendorfer et al., 2011). The average scores (velocity and distance) of the two scored TGMD-3 trials were retained for the analysis of the four skills on which product measures were collected. The hop and jump distances were calculated in centimeters. The maximum throw and kick velocities were measured with a radar gun (Stalker, Inc.; Stodden et al., 2006a, 2006b, 2014).

Data Analysis

Procedures for data processing included (a) cleaning and entering data into Microsoft Excel, (b) rechecking for correct entry, (c) saving to a password protected computer, and (d) using SPSS Version 25 statistical software (Armonk, NY) to complete all analyses.

Descriptive statistics were calculated reported for all participants ($N = 45$) and included age, biological sex, race/ethnicity, degree of vision, and body mass index. Body mass index (BMI) was calculated with participant height (inches) and weight (pounds; United States Department of Health and Human Services, 2010). Mean and standard deviations were calculated for raw TGMD-3 scores (total, ball skills, and locomotor skills), product scores by skill, perceived motor competence, PedsQLTM, and physical activity. Raw total and sub scale scores were used to analyze motor competence process (TGMD-3) and HRQoL (PedsQLTM). Average scores were used for analysis of perceived motor competence (by item) and physical activity (total steps-per-day). For a total product score, scores from each skill (hop, jump, kick, and throw) were converted to z-scores and combined for further analysis (Stodden et al., 2009). To further analyze motor competence with a comprehensive motor competence score (process and product), TGMD-3 total

scores were converted to z-scores and combined with the total product z-scores. Two separate four (VI classifications) by three group (VISC, VI, and healthy peers) ANCOVAs were calculated to explore mean differences for motor competence and physical activity with the presence of a co-morbidity as a covariate. Then bonferroni post hoc analyses were conducted to determine any differences between groups. To examine group differences for HRQoL and perceived motor competence, a Kruskal Wallis Test with three groups (VISC, VI, and healthy peers) was used, followed by post hoc analyses using Mann Whitney Tests.

Results

For the purposes of this study, descriptive statistics for motor competence, physical activity, perceived motor competence, and HRQoL were conducted by participant group (VI, VISC, and comparison; See Table 3.2).

Table 3.2. Descriptive Statistics for Variables of Interest by Group

| | Total | VISC | VI | Comparison |
|---------------|-----------------------|-----------------------|-----------------------|----------------------|
| | M(SD) | M(SD) | M(SD) | M(SD) |
| TGMD-3 | 65.69(19.37) | 58.53(20.59) | 56.40(15.67) | 82.13(8.52) |
| TGMD-3 Loc. | 31.81(7.60) | 30.33(9.16) | 28.80(6.53) | 36.27(4.65) |
| TGMD-3 Ball | 33.89(13.31) | 28.20(13.23) | 27.60(10.86) | 45.87(5.74) |
| Combined MC | .00(2.85) | -1.06(2.74) | -1.58(1.78) | 2.64(1.88) |
| Product MC | .00(1.93) | -.69(1.74) | -1.10(1.06) | 1.79(1.52) |
| Total | | | | |
| Product Ball | 56.40(29.50) | 45.83(25.24) | 39.57(19.26) | 83.80(22.37) |
| Product Loc. | 408.44(159.71) | 354.88(156.52) | 324.39(92.62) | 546.04(127.13) |
| Average Steps | 14813.64 (6256.95) | 17524.47 (5330.37) | 17364.33 (4167.96) | 9552.13 (6256.95) |
| Perceived MC | 3.07(.78) | 2.82(.88) | 2.99(.74) | 3.4(.63) |
| HRQoL Total | 78.70(13.03) | 78.33(16.89) | 81.77(13.26) | 76.00(7.37) |
| Emotional | 294.44(79.04) | 59.67(17.57) | 63.67(14.20) | 53.33(14.72) |
| Physical | 83.56(16.60) | 78.00(21.45) | 87.33(13.61) | 85.33(13.02) |
| Social | 82.41(17.96) | 81.67(21.41) | 83.89(18.76) | 81.67(14.16) |
| Mental | 73.52(18.14) | 80.56(16.57) | 72.78(22.38) | 67.22(12.78) |

MC = motor competence

Loc. = locomotor

Multiple separate One-Way Analysis of Covariance (ANCOVA) with three groups (VI, VISC, and comparison group) were conducted to examine the group differences in motor competence (TGMD-3, product measures, and a combination of process/product) and physical activity (average steps) above and beyond the presence of a co-morbidity. The ANCOVAs revealed a significant difference between groups for motor competence and physical activity (see Table 3.3). The first dependent variable examined was the motor competence product total combined z-scores ($F = 14.51, p > .001, \eta_p^2 = .41$), ball skills subscale ($F = 14.55, p < .001, \eta_p^2 = .42$), and locomotor subscale ($F = 11.21, p < .001, \eta_p^2 = .35$). Bonferroni post hoc analyses revealed significantly higher for motor competence combined product z-scores for the comparison group over both the VISC group (+ 2.35, $p < .001$), and the VI group (+ 2.81, $p < .001$). However, the VI and VISC groups were not significantly different from one another (VISC group scores higher than VI, + .46, $p > .05$). Pairwise comparison results from the Bonferroni post hoc analyses indicated significantly higher scores for the comparison group than the VISC group for motor competence product scores ball skills total velocity (+ 35.91, $p < .001$), and product locomotor total distance (+ 181.19, $p < .001$). Bonferroni post hoc analyses also indicated significant higher scores for the comparison group than the VISC group for motor competence product scores ball skills total velocity (+ 42.99, $p < .001$), and product locomotor total distance (+ 215.67, $p < .001$). Although the differences between the VISC group and the VI group were not significant, for motor competence product scores, the VISC group means were above the VI group means (ball skills total velocity + 7.09, $p > .05$; locomotor total distance + 34.48, $p > .05$). The second, dependent variable examined was physical activity, which was measured by average steps ($F = 11.03, p < .001, \eta_p^2 = .35$). The Bonferroni post hoc analyses results

indicated significantly lower average steps per-day for the comparison group than VISC participants (- 8156.32, $p < .001$) and VI participants (- 7922.59, $p < .001$). A post-hoc priori power analysis was conducted using G*Power Software version 3.1.9.4 for an ANCOVA to ensure a type 2 error was not made. To calculate, an alpha level of 0.05, an η_p^2 of .35 and .42, participants ($N = 45$), three groups, and one covariate. Based on this calculation we had more than sufficient power ($1 - \beta$ err prob .999) to run multiple ANCOVAs with three groups and one covariate.

Table 3.3. ANCOVA Group Differences

| | <i>M(SD)</i> | SS | DF | F | Sig. |
|---------------------|-------------------|------------|-----------|-------------|-------------|
| Product MC Total | .00(1.93) | 63.14 | 2 | 14.51 | .00** |
| Product Locomotor | 408.44(159.71) | 373225.13 | 2 | 11.21 | .00** |
| Product Ball Skills | 56.40(29.50) | 14785.24 | 2 | 14.55 | .00** |
| Combined MC | .00(2.85) | 137.37 | 2 | 14.33 | .00** |
| Physical activity | 14813.64(6256.95) | 623071051. | 2 | 311535525.7 | .00** |
| | | 51 | | 6 | |

Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively. MC = motor competence

Group differences for the TGMD-3 were analyzed with three separate Kruskal Wallis analyses total scores ($H = 16.63, p < .001$), ball skills subscale ($H = 18.63, p < .001$), and locomotor subscale ($H = 8.12, p = .017$). The results from the Mann-Whitney test post hoc analyses indicated that participants in the comparison group achieved TGMD-3 scores that were significantly higher than the participants' scores in the VISC group (total scores $U = 38.5, p = .002$; ball skills $U = 30.5, p < .001$), except in locomotor skills ($U = 73.0, p = .136$). The comparison group TGMD-3 scores were also significantly higher than participants in the VI group (total scores $U = 18.0, p < .001$; ball skills $U = 16.0, p < .001$; and locomotor $U = 40.5, p = .002$). However, differences in the participant TGMD-3 scores between the VI and VISC groups were not significantly different.

To examine group differences for HRQoL and perceived motor competence, due to the nonparametric nature of the data, two separate Kruskal Wallis analyses were used. The Kruskal Wallis test revealed that the groups were not statistically different in HRQoL ($H = 2.10, p > .05$) or perceived motor competence ($H = 4.44, p > .05$; See Table 3.4). Although, the asymptotic relative efficiency of the Kruskal Wallis is .955 (Marascuilo & McSweeney, 1977), there is still a possibility of inadequate power due the small sample size that may have contributed to no statistical difference being detected (See Figure 3.1 and Figure 3.2). Due to the difference displayed in Figure 3.2 a Mann-Whitney test post hoc analysis for perceived motor competence average scores was run between the VISC and comparison group, and indicated that participants in the VISC group had significantly lower perceived motor competence ($U = 65.0, p = .039$).

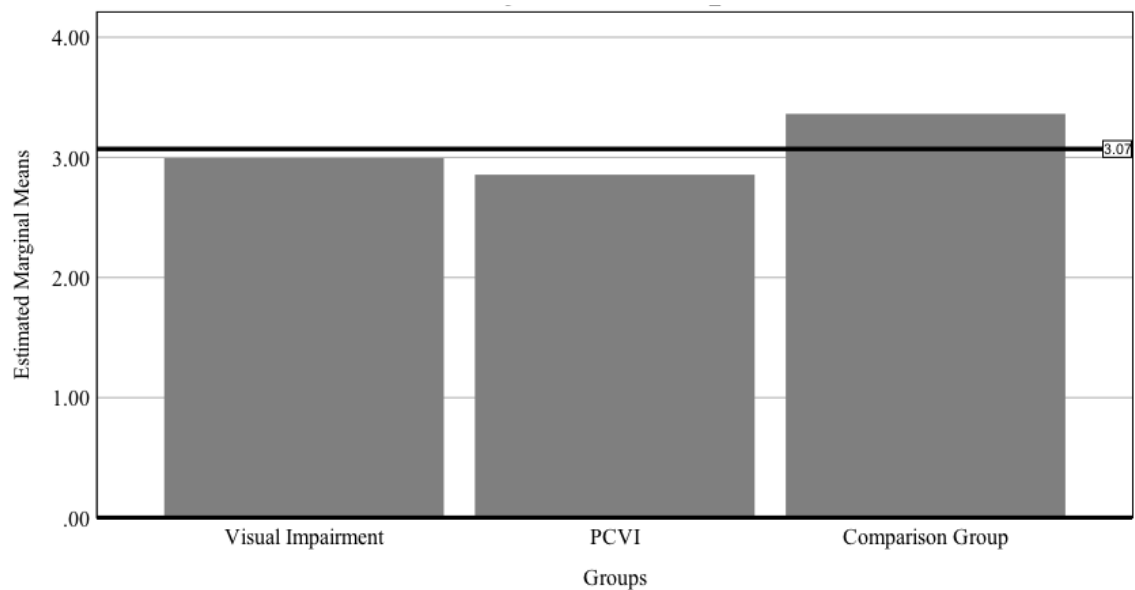
Table 3.4. Kruskal Wallis Group Differences

| | <i>M(SD)</i> | DF | H | Sig. |
|----------------------------|--------------|-----------|----------|-------------|
| TGMD-3 | 65.69(19.37) | 2 | 16.63 | .00** |
| TGMD-3 Locomotor | 31.80(7.60) | 2 | 8.12 | .02* |
| TGMD-3 Ball Skills | 33.89(13.31) | 2 | 18.63 | .00** |
| Perceived Motor competence | 3.07(.78) | 2 | 4.44 | .11 |
| HRQoL | 78.70(13.03) | 2 | 2.10 | .35 |

Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively.

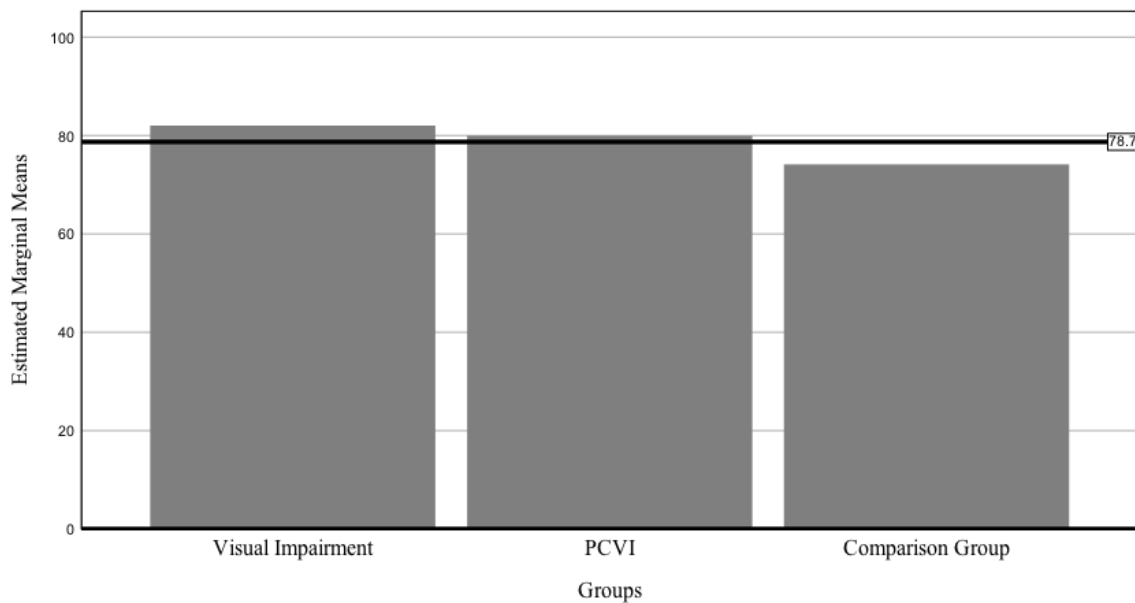
Discussion

The purpose of this study was to examine group differences between youth with a VI, VISC, and a healthy comparison group regarding HRQoL, motor competence, physical activity, and perceived motor competence. The results of this study indicated that both the VI and VISC groups were significantly below their comparison group in their process and product scores of motor competence. The product measures of motor competence appeared to be more sensitive in differentiating participant's locomotor skills, whereas TGMD-3



Covariates appearing in the model are evaluated at the following values: Multiple_Dis=1 = .24

Figure 3.1. Estimated Means of Average Perceived Motor Competence Scores.



Covariates appearing in the model are evaluated at the following values: Multiple_Dis=1 = .24

Figure 3.2 Means of Average Perceived HRQoL Scores

differences were not significant between groups and product total locomotor scores were significant. This finding aligns with the recommendations of other researchers in the field to use a combination approach, particularly with such a large age span, to differentiate between motor competence performance levels more effectively across the lifespan (Burton & Miller, 1998; Hands, 2002; True et al., 2017).

The participants in the VISC group and VI group were not statistically different from one another in motor competence (process or product), which differed from our initial hypothesis. However, it is possible that the participants age at onset of the VI was a stronger indicator than the previous cancer diagnosis. Although, the difference was not significant the VISC group mean scores were higher on both product (locomotor and ball skills) and process (locomotor and ball skills), which most of the participants in the VI group have had a VI since birth. The difference between acquired and congenital VI has not been addressed in the literature and should be examined in the future to determine the impact this may have on motor competence for youth with a VI.

An unexpected finding of this study is that comparison group's mean HRQoL was much lower than both the VI and VISC groups. This finding is contradictory to previous literature for individuals with a pediatric cancer, VI, or chronic illness in comparison to their healthy peers (Boulton et. al., 2006; Chadha & Subramanian, 2011; Chen et al., 2005; Gohar et al., 2011; Habib & Irshad, 2018; Ness et al., 2015; Wong et al., 2009; Wright et al., 2005). Due to this finding, future research should measure HRQoL in low socioeconomic rural areas with a larger sample of youth in the general population. The relationship between socioeconomic status and HRQoL has been identified as significant in adolescent immigrant students and youth with heart disease (Cassedy et al., 2013; Chau

et al., 2012). Previous studies have reported a mean score for healthy youth using the PedsQL™ total score between 81 and 86, whereas our sample mean was much lower with a total score of 76 (Chan et al., 2005; Chen et al., 2005; Lam et al., 2013).

Additionally, the mean average steps for both the VI and VISC groups were significantly higher than the comparison group, which is in contrast to the previous literature (Haegele & Porretta, 2015). However, this difference may have been due to the VI and VISC groups wearing the Fitbits™ during a week-long summer sports camp, and the comparison group wearing their Fitbits™ during a typical school week. Consequently, participants at the school could choose to be physically active in their own time, whereas participants at the sports camps participated in scheduled physical activity throughout the day. Although a previous study found a moderate association ($r = .35$) with participants physical activity recall and physical activity at a sports camp for the blind and VI (Brian et al., 2019a).

There were several limitations in this study. First, due to the nature of the population, only an small sample size could be obtained via a purposive convenience sample. Similar studies on youth with VIs or VISC have had small samples as well ranging from 14 to 46 participants due to the low incidence population (Colgan et al., 2016; Haegele et al., 2017; Lieberman & McHugh, 2001). However, due to the small sample in the present study, the results should be interpreted as preliminary. Second, the participants' physical activity levels may have been strongly influenced by the testing site (school or a sports camp), the setting of this study was necessary due to the limited access to larger groups of these low incidence populations. Third, multiple self-report surveys were used to collect the data for perceived motor competence and HRQoL, possibly contributing to biased

responses due to the nature of self-reporting. Finally, although we controlled for a co-morbidity we don't know if the co-morbidity was a direct link to the cancer or treatment as well and should be examined in the future.

Future research in this area should examine differences between these populations with a larger sample. Although, in significant HRQoL and perceived motor competence may be trending towards a difference between VI and VISC groups (See Figure 3.1 and 3.2). However, the sample may not have been large enough to detect these differences in HRQoL or perceived motor competence and should be examined in the future with a larger sample. There remain many gaps in the literature, particularly correlational and predictive studies on youth with VIs and VISC. These studies are needed to address questions related to how motor competence, physical activity, perceived motor competence, and HRQoL relate to each other. Additionally, research is needed to test the assertions of Stodden and colleagues' (2008) regarding relationships between motor competence, perceived motor competence, and physical activity in this population.

In conclusion, the motor competence of youth with VIs and a VISC have a tendency to be lower than peers without a VI or VISC, but are not different from each other. Based on the findings we recommend that physical educators emphasize motor skill development tailored to youth with VIs and VISCs, with particular focus on ball skills which were even lower than locomotor skills. Additionally, the presence of a co-morbidity should not be overlooked when planning research or instruction. Future research in this area is necessary to replicate the group differences found and expand by further examination of the relationships between motor competence, physical activity, perceived motor competence, and HRQoL for these populations.

CHAPTER 4:
STUDY 3 UNDERLYING MECHANISMS PREDICTING PHYSICAL ACTIVITY
AND HRQOL IN YOUTH WITH VISUAL IMPAIRMENTS WITH AND
WITHOUT CANCER

Children that have a disability are significantly more likely to be overweight than their same aged peers without a disability, and this tends to become more significant as children reach adolescence (CDCP, 2020; De et al., 2008). Specifically, youth with a VI are more likely to be overweight or obese than their sighted peers (Lieberman et al., 2010; Lieberman & McHugh, 2001; Weil et al., 2002). An unhealthy weight status can lead to life-long negative health (Quatman, Ford, Myer, & Hewett, 2006) and social-emotional impacts (Taunton et al., 2018), which can then compound with the potential social impacts of having a VI or VISC. Physical activity has been identified as an important predictor and intervention mechanism for an unhealthy body weight status (e.g., obesity) in the general population (Physical Activity Guidelines Advisory Committee, 2018). Additional benefits associated with physically activity levels include metabolic and bone health (Physical Activity Guidelines Advisory Committee, 2018), social benefits (e.g., temperament; Taunton et al., 2018), and psychological benefits (e.g., self-esteem; Columna et al., 2015).

The implications of low physical activity, unhealthy weight status, and a VI may be factors in the increased risk of low HRQoL for individuals with VI or VISC. HRQoL is a multidimensional construct of one's perception regarding their position in life, based on

their environmental and individual contexts (WHO, 2011). The three dimensions of HRQoL include physical, psychological, and social well-being and function (Matza et al., 2004; Ravens-Sieberer et al., 2006). The three dimensions of HRQoL encompass: an individual's perception of daily physical activities (physical dimension), self-concept and self-esteem among other psychological variables (emotional dimension), and social relationships including family and friends (social functioning; Bowling, 1999; Kaplan et al., 1993). Up to this point, little has been done to examine HRQoL from a physical dimension perspective for youth with a VI. Much of the literature on HRQoL in individuals with VIs has focused on the social emotional aspects and quality of life in the workplace or school. Young adults with VIs have been found to have a low HRQoL in comparison to their sighted peers (Elsman et al., 2017). However, the etiology of the VI (e.g., nystagmus) impacted the degree of negative implications on HRQoL, with youth with a visual pathway impairment having the lowest HRQoL (Boulton et al., 2006). Additionally, there are many important factors that have been positively associated with an increased quality of life for individuals with VIs including: independent travel, socialization, relaxation, sense of independence, and involvement in recreational physical activity (Ball & Nicolle, 2015; Columna et al., 2015). Factors of the physical dimension (physical activity, motor competence) have often been overlooked as potential predictors of HRQoL, particularly for youths with a VI.

To theoretically guide this study, a model designed by Stodden and colleagues (2008) was used to examine physical activity levels and HRQoL from a physical perspective for youth with a VI or VISC. The 2008 conceptual model by Stodden and colleagues conceptualizes the relationship between motor competence and physical

activity engagement or disengagement, mediated by perceived motor competence and health-related fitness. Based on this model, engagement or disengagement in physical activity was used to predict individuals' body weight status (Stodden et al., 2008). However, we further hypothesized that this conceptual model would also predict HRQoL for youth with a VI.

Youth with a VI are more likely to have lower motor competence, perceived motor competence, physical activity levels, and higher rates of obesity (Brian et al., 2016; Haegele et al., 2015; Houwen et al., 2007; Wagner et al., 2013). Moreover, a VISC (e.g., brain tumor, retinoblastoma) may further exacerbate the likelihood of negative effects on motor competence, physical activity, and perceived motor competence identified in the pediatric cancer and VI populations (Augestad & Jiang, 2015; Brian et al., 2016; Florin et al., 2007; Green et al., 2013; Haegele et al., 2015; Hartman et al., 2010; Hopkins et al., 1987; Houwen et al., 2007; Leone et al., 2014; Ness et al., 2015; Piscione et al., 2014; van Brussel et al., 2005; Wagner et al., 2013; Weil et al., 2002). In addition to difficulties in the physical domain youth with a VI often have a higher chance of a co-morbidity with over 50% of individuals with VIs also have at least one other impairments (Packer & Kirchner, 1985).

The implications of low motor competence, perceived motor competence and physical activity may lead to a negative spiral of disengagement in physical activity and a negative spiral towards low HRQoL (Stodden et al., 2008). Yet, much remains unknown for youth who have a VI or a VISC about the associations among motor competence, perceived motor competence, and physical activity. Additionally, little is known about how these variables interact with overall HRQoL in these populations of youth. Therefore, the

purpose of this study was to examine the underlying mechanisms predicting physical activity and HRQoL for youth with a VI or VISC.

Research Question 1 Which of the factors (motor competence and perceived motor competence) predict physical activity in youth with a VI and VISC after controlling for significantly correlated (to physical activity) participant demographics?

Hypothesis 1. Motor competence and perceived motor competence will predict physical activity for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to physical activity).

Research Question 2. Which of the factors (motor competence, perceived motor competence, and physical activity) predict body weight status in youth with VI and VISC after controlling for significantly correlated (to BMI) participant demographics?

Hypothesis 2. Motor competence, perceived motor competence and physical activity will predict body weight status for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to BMI).

Research Question 3. Which of the factors (motor competence, perceived motor competence, and physical activity) predict HRQoL in youth with VI and VISC after controlling for significantly correlated (to HRQoL) participant demographics?

Hypothesis 3. Motor competence, perceived motor competence and physical activity will predict HRQoL for all groups (VI and VISC), after controlling for participant demographics (that are significantly correlated to HRQoL).

Methodology

Participants and Setting

A purposive convenience sample of youth (7-18 years old) were recruited from summer camps for children who are blind and visually impaired. These camps were located in the following states: New York, Delaware, Texas, and Florida. All participants with VISC were included in this study, and participants from the VI group were matched by age, biological sex, and degree of vision. Degree of vision (severity) is most commonly classified with visual acuity, visual field, and contrast sensitivity (American Printing House for the Blind, 2016; NASEM, 2016). For the purposes of this study, degree of vision will be categorized based on an established sport-based classification system, B1 (e.g., no functional vision) to B4 (e.g., low vision), in this system all classifications are based on the best eye including eye correction (USABA, 2017). Specifically, B1 classification is the lowest degree of vision (e.g., includes no light perception to light perception), B2 classification (e.g., visual acuity up to 20/600), B3 classification (visual acuity between 20/600–20/200 and a visual field between 20 – 5 degrees), and B4 classification is considered the highest degree of vision (e.g., visual acuity between 20/200 – 20/70 and a visual field greater than 20 degrees; USABA, 2017).

A total of 30 participants were included in this study in two groups, VISC ($n = 15$) and VI ($n = 15$) with a mean age of 12.33 ± 2.64 years, biological sex (male = 18, female = 12), race/ethnicity (82.8% Caucasian, 6.9% Asian, 6.9% Black, 3.4% other), and body mass index ($M_{BMI} = 21.38 \pm 5.77$; see Table 4.1). Some of the participants in this study also had a co-morbidity ($n = 10$). The specific diagnoses of the co-morbidity ($n = 1$ participant had two) were as follows: other health impairment ($n = 3$), hearing impairment

($n = 2$), traumatic brain injury ($n = 2$), emotional disturbance ($n = 1$), developmental delay ($n = 1$), autism spectrum disorder ($n = 1$), and multiple disabilities ($n = 1$). The majority of the participants VIs were congenital ($n = 17$), with the rest being acquired VIs ($n = 13$).

Table 4.1. Descriptive Statistics for Demographic Variables by Group

| | VISC | | VI | |
|------------------|-------------|------|-------------|------|
| | M(SD) | | M(SD) | |
| Age (years) | 12.47(2.85) | | 12.20(2.51) | |
| | <i>N</i> | % | <i>N</i> | % |
| Biological Sex | 9(Male) | 60.0 | 9(Male) | 60.0 |
| Co-Morbidity | 6 | 40.0 | 4 | 26.7 |
| Congenital VI | 5 | 33.3 | 12 | 80.0 |
| Degree of Vision | | | | |
| B1 | 7 | 46.7 | 8 | 53.3 |
| B2 | 4 | 26.7 | 3 | 20.0 |
| B3 | 2 | 13.3 | 2 | 13.3 |
| B4 | 2 | 13.3 | 2 | 13.3 |

Instrumentation

Health-related quality of life. HRQoL was measured using the Pediatric Quality of Life Inventory™ Version 4.0 Short Form 15 (PedsQL™ 4.0 SF15) survey (Varni et al., 1998). Three different age bands of the PedsQL™ 4.0 SF15 – SF were used including: Teen Report 13 – 18, Child Report 8 – 12, and Young Child Report 5 – 7. Each of the age bands are comprised of 15 items and four dimensions. The four dimensions include physical functioning (5-item), emotional functioning (4-item), social functioning (3-item), and school functioning (3-item; Varni et al., 1998). The participants responded to each prompt on a five-option lykert scale, 0 (never) to four (almost always; Chen et al., 2005). Surveys are scored by reverse scoring and transforming each item onto a scale of zero to 100 (e.g., 0 = 100, 4 = 0), then dividing by the number of items in the dimension; a higher score indicates a superior perception of HRQoL (Varni et al., 1998, 1999). The PedsQL™ 4.0 SF15 – SF Generic Core Scale results have demonstrated acceptable validity and

reliability for pediatric HRQoL to evaluate internal consistency Cronbach's Alpha was calculated as total score ($\alpha = .82$), physical health ($\alpha = .60$), emotional functioning ($\alpha = .76$), social functioning ($\alpha = .74$), and school functioning ($\alpha = .75$; Chen et al., 2005).

Motor competence. The Test of Gross Motor Development third edition (TGMD-3) was used to measure process motor competence including ball skills and locomotor subscales (Webster & Ulrich, 2017). The locomotor subscales include the following skills: run, hop, skip, gallop, jump, and slide (Webster & Ulrich, 2017). The ball skills subscale includes: two-handed strike, one-handed strike, one-handed dribble, two-handed catch, kick, overhand throw, and underhand throw (Webster & Ulrich, 2017). All scored trials are coded with either a one (met criteria) or zero (did not meet the criteria). The validity and reliability psychometrics using the TGMD-3 are strong for youth with VIs, up to 19 years of age (Brian et al., 2018). Modifications specifically for individuals with VIs were made for implementing the TGMD-3, such as utilizing a beep kickball and a beep baseball (Brian et al., 2018). According to Webster and Ulrich (2017) the TGMD-3 can be used as a criterion reference for all ages, assuming an individual does not earn a perfect score.

Product scores were collected on the following TGMD-3 motor skills: hop (distance), jump (distance), kick (velocity), and throw (velocity; Bürgi et al., 2011; True et al., 2017; Stodden et al., 2014). To measure the velocity product outcomes maximum miles per hour for throw and kick using a Stalker radar gun was recorded (Stalker Radar, Plano, TX; Stodden et al., 2014). Participants are asked to hop from cone to cone (5 meters apart), for two scored trials. Three consecutive hops total distance (heel-to-heel) in centimeters were recorded as the product outcome for each trial using video analysis software (Dartfish Video Analysis Software, Version 7.0). Standing long jump is to be measured for two trials

from the takeoff line to the back of the closest heel on landing, which was recorded onsite with a tape measure to the nearest centimeter (Stodden et al., 2014).

Perceived motor competence. The Test of Perceived Motor Competence for children with VIs (TPMC-VI; ages 7 to 8; Brian et al., 2016), Self-Perception Profile for Children (ages 9 to 13 years; Harter, 2012b), or the Self-Perception Profile for Adolescents (ages 14 to 19 years; Harter, 2012a) were used in this study to measure perceived motor competence in all participants (Brian et al., 2016, 2018). The TPMC-VI is a modified version of the Pictorial Scale of Perceived Competence and Social Acceptance, utilizing vignettes to describe the content in the photos for youths with a VI (Brian et al., 2016; Harter & Pike, 1984). The TPMC-VI is a six-item questionnaire with a two-question, forced-choice response, based on a specific motor skill situation (e.g., “really true for me” or “sort of true for me”). The scoring for this measure is a scale from one to four, with one being low and four being high. The Self-Perception Profile for Children and the Self-Perception Profile for Adolescents follow the same format as the TPMC-VI except that there are only five items on the adolescent scale. All three measures should require between five to ten minutes to complete. The psychometric properties of TPMC-VI reveal strong content and face validity and reliability for individuals with a VI (Brian et al., 2016). Both of the batteries Self-Perception Profile for Children and the Self-Perception Profile for Adolescents have moderate to strong psychometric properties and consistently produce results that are considered valid and reliable, (Harter, 2012a, 2012b).

Physical activity. Physical activity data were collected using Fitbit Inspire™ and Fitbit Ace2™ activity trackers. The Fitbit Inspire™ is for use with children 13 years of age and older wore the and the Fitbit Ace2™ is for children 12 years of age and younger. The

Fitbit Inspire™ and Fitbit Ace2™ wearable activity trackers have a sensor that uses a tri-axis accelerometer to measure steps taken, distance traveled, calories burned, active minutes, hourly activity and stationary time (Fitbit, San Francisco, CA). Additionally, the Fitbit Inspire™ and Fitbit Ace2™ devices are capable of submersion in water up to 50 meters, allowing participants to wear them during aquatic activities. The Fitbit devices store seven days' worth of minute-by-minute detailed motion data that can later be downloaded for analysis (Fitbit, San Francisco, CA).

Body Weight Status. To measure participants' body weight status actual standing height and weight were measured using a portable stadiometer and digital scale. Then BMI was calculated with participant height (inches) and weight (pounds; United States Department of Health and Human Services, 2010).

Procedures

All assessments for individuals with a VI and VISC were administered in the months of June, July, and August 2019. Each of the participants with VISC were matched by age, degree of vision, and biological sex with a participant in the VI group. The assessments were conducted in a gymnasium, open room, or a designated outdoor space at the summer camps. The assessments were administered in the same order at each camp and school district to ensure assessment order was not a factor in the results. University Institutional Review Board (IRB) approval and approval from external entities (camps and school districts) was obtained prior to data collection. Furthermore, all rules, regulations, and training requirements were observed throughout the project. Upon arrival to camp, parents and participants completed parental consent and child assent before being enrolled in the study.

Then, participants were assigned and provided a Fitbit Inspire™ or Fitbit Ace2™ to collect their physical activity data throughout the week. Participants were instructed to wear the Fitbit Inspire™ or Fitbit Ace2™, on their non-dominant hand, for four to five days, for a minimum of ten hours per day (Pate et al., 2004; Puyau et al., 2002). The span of days the activity tracker was worn was dependent on whether data was collected from a participant at a summer camp, and the duration of that camp. The Fitbit Inspire™ and Fitbit Ace2™ devices are capable of submersion in water up to 50 meters, allowing participants to wear them during aquatic activities, which children assessed at each of the camps participated in daily.

After being assigned a Fitbit Inspire™ or Fitbit Ace2™, participants' height and weight were measured while parents completed the demographic questionnaire. Participants' actual standing height and weight were measured using a portable stadiometer and digital scale. Demographic information was collected using a questionnaire, completed by the participants parent/guardian, that included participants' birth date, height, weight, race/ethnicity, biological sex, degree of vision, self-reported VI diagnosis, self-reported cancer diagnosis, and age at onset of VI and/or cancer diagnosis.

Next, each participant completed a measurement of perceived motor competence (TPMC-VI, Self-Perception Profile for Children, or Self-Perception Profile for Adolescents) and HRQoL (PedsQL™ 4.0 SF15). Each participant was provided the appropriate age band of each assessment for perceived motor competence and HRQoL. The participants were then asked to answer the prompts, either one-on-one with a trained coach or on independently (large print and braille copies were provided). Each survey took between five to fifteen minutes to complete.

The final assessments administered were the motor competence measurements (TGMD-3 and product scores). All motor competence measurements were video recorded for coding purposes. Before data coding began interrater reliability was calculated using an intraclass correlation coefficient for ball skills subscale ($ICC = .968$, $95\% CI = .94 - .99$, $p < .001$) and locomotor subscale ($ICC = .949$, $95\% CI = .90 - .98$, $p < .001$) to demonstrate agreement between both of the coders. For each gross motor skill, the equipment, preparation and protocol from the TGMD-3 (Webster & Ulrich, 2017) were used, except for modifications for VI used from Brian and colleagues (2018). For each skill, demonstration and verbal instructions were provided. The product data was collected on the same trials following the TGMD-3 protocol. Participants were instructed to perform their best (e.g., “Jump as far as you can”), to facilitate them putting forth true effort into their attempt at each motor skill (Langendorfer et al., 2011). The average scores (velocity and distance) of the two scored TGMD-3 trials were retained for the analysis of all four skills product measures. The hop and jump distances were calculated in centimeters. The maximum throw and kick velocities (miles per hour) were measured with a radar gun (Stalker, Inc.; Stodden et al., 2006a, 2006b, 2014).

Data Analysis

Procedures for data processing included (a) cleaning and entering data into Microsoft Excel, (b) rechecking for correct entry, (c) saving to a password protected computer, and (d) using SPSS Version 25 statistical software (Armonk, NY) to complete all analyses.

A mean and standard deviation of descriptive statistics were reported for all participants with a VISC ($N = 15$) or VI ($N = 15$) including age, biological sex,

race/ethnicity, degree of vision, and body mass index (BMI). Mean and standard deviations were calculated for raw TGMD-3 scores (total, ball skills, and locomotor skills), product scores by skill, perceived motor competence, PedsQL™, and physical activity. Raw total and sub scale scores were used to analyze motor competence process (TGMD-3) and HRQoL (PedsQL™). Average scores were used for analysis of perceived motor competence (by item) and physical activity (total steps-per-day). To analyze motor competence product scores for locomotor and ball skills, a sum of total average distance steps taken per day and for ball skills total average miles per hour were used. For a total product score, scores from each skill (hop, jump, kick, and throw) were converted to z-scores and combined for further analysis (Stodden et al., 2009). To examine the relationships between the variables and participant demographics, Pearson's two-tailed bivariate correlations were conducted. For interpretation of the correlations 0.30 to 0.50 were identified as a moderate strength and correlations 0.60 and greater were as identified as strong (Lomax & Hahs-Vaughn, 2013). Then, 3 separate three and four -level Hierarchical Regressions with forced entry were used to predict physical activity, body weight status, and HRQoL after controlling for significantly correlated participant demographics.

Results

The purpose of this study was to examine the underlying mechanisms predicting physical activity, body weight status, and HRQoL for youth with a VI or VISC, above and beyond significantly correlated demographics (e.g., etiology). See Table 4.2 for descriptive statistics.

Table 4.2. Descriptive Statistics for Variables of Interest by Group

| | VISC | Visual Impairment |
|------------------|-------------------|-------------------|
| | M(SD) | M(SD) |
| TGMD-3 | 58.53(20.59) | 56.40(15.67) |
| Product MC Total | .75(.75) | .47(.53) |
| Average Steps | 17524.47(5330.37) | 17364.33(4167.96) |
| Body mass index | 22.67(6.07) | 20.08(5.33) |
| HRQoL Total | 78.33(16.89) | 81.77(13.27) |

Next, to answer the research questions, Pearson's two-tailed bivariate correlations were conducted to determine what participant demographics and variables were significantly associated with physical activity and HRQoL. Physical activity (average steps) had a strong and positive association with product motor competence ($r = .71, p < .001$), TGMD-3 ($r = .60, p < .001$), and participants degree of vision ($r = .58, p < .001$). Body weight status (BMI) was only significantly associated with participants years of age ($r = .55, p < .001$). Additionally, physical activity had a moderate correlation with HRQoL ($r = .42, p = .020$). HRQoL (PedsQL™) had a strong and positive association with product motor competence ($r = .53, p = .003$) and a moderate correlation with perceived motor competence ($r = .42, p = .020$), TGMD-3 ($r = .37, p = .047$), physical activity ($r = .42, p = .020$), biological sex ($r = -.37, p = .043$), and a co-morbidity ($r = -.42, p = .022$). For the complete correlation matrix see Table 4.3.

Following the Pearson's correlations, a three-level hierarchical regression with forced entry was used to predict physical activity for youth with a VI and VISC. Level 1 accounted for degree of vision, level 2 accounted for perceived motor competence, and level 3 included total product/process motor competence scores. For model three, level 1 degree of vision ($\beta = .31, p = .056$), level 2 perceived motor competence ($\beta = .09, p = .53$), and level 3 motor competence ($\beta = .49, p = .007$) predicting participants average step count

Table 4.3. Two-tailed Bivariate Correlation Matrix

| | Age | Bio. Sex | BMI | Degree of Vision | Co- Morbidity | PMC | TGMD-3 | Product MC | Steps | Etiology | HRQoL |
|---------------------|-------|-------------|------|------------------------|------------------|------|--------|------------|-------|----------|-------|
| Age | 1.00 | | | | | | | | | | |
| Bio. Sex | -.34 | 1.00 | | | | | | | | | |
| BMI | .55** | -.22 | 1.00 | | | | | | | | |
| Degree of Vision | .27 | .20 | .07 | 1.00 | | | | | | | |
| Co-Morbidity | -.12 | .14 | .01 | .20 | 1.00 | | | | | | |
| PMC | .03 | -.28 | .06 | .16 | -.21 | 1.00 | | | | | |
| TGMD-3 | .50** | -.30 | .07 | .54** | -.05 | .45* | 1.00 | | | | |
| Product MC | .63** | -.29 | .18 | .47** | -.13 | .37 | .86** | 1.00 | | | |
| Steps | .32 | -.08 | -.11 | .58** | -.06 | .34 | .60** | .71** | 1.00 | | |
| Etiology | .10 | -.12 | .25 | .08 | .16 | .00 | .13 | .20 | .01 | 1.00 | |
| HRQoL | .25 | -.37* | -.09 | .07 | -.42* | .42* | .37* | .53** | .42* | -.07 | 1.00 |

Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively.

Bio. Sex = biological sex

PMC = perceived motor competence

MC = motor competence

scores. Model three accounted for and 54% of the variance explained ($R^2 = .54, p = .007$) in participants average step count scores. Motor competence specifically added 15% of variance explained ($R^2 \Delta = .14, p = .007$) in participants average step count scores beyond degree of vision and perceived motor competence. A post-hoc priori power analysis was conducted using G*Power Software version 3.1.9.4 for a three-level hierarchical regression by imputing an alpha level of 0.05, observed $R^2 = .54$, participants ($N = 30$), two measured predictors, with three total predictors ($1 - \beta$ err prob = .99). See Table 4.4. for the complete results of the three-level hierarchical regression.

Then a four-level hierarchical regression with forced entry was used to predict body weight status for youth with a VI and VISC. Level 1 accounted for years of age, level 2 accounted for perceived motor competence, level 3 included TGMD-3 total scores and total product motor competence scores, and level 4 included physical activity average steps. Years of age was the only significant predictor of body weight status (BMI) accounting for 33% of the variance in participant BMI ($\beta = .55, p = .002$). None of the other models significantly added to predicting BMI, see Table 4.5. for the complete results of the four-level hierarchical regression. A post-hoc priori power analysis was conducted using G*Power Software version 3.1.9.4 for a four-level hierarchical regression by imputing an alpha level of 0.05, observed $R^2 = .33$, participants ($N = 30$), three measured predictors, with four total predictors ($1 - \beta$ err prob = .80).

To examine the underlying mechanisms predicting HRQoL a four-level hierarchical regression with forced entry was used for youth with a VI and VISC. Level 1 accounted for a co-morbidity and biological sex, level 2 accounted for perceived motor competence, level 3 included motor competence (product and process), and level 4

Table 4.4. Factors Predicting Physical Activity

| | Model 1 | | | Model 2 | | | Model 3 | | |
|------------------------------------------|----------------|-----------|---------------------------|----------------|-----------|---------------------------|----------------|-----------|---------------------------|
| | B | SE | β | B | SE | β | B | SE | β |
| (Constant) | 12736.06 | 1449.21 | | 8745.36 | 2757.37 | | 14674.85 | 3180.43 | |
| Degree of Vision | 2478.08 | 663.78 | .58** | 2304.04 | 651.39 | .54** | 1335.12 | 665.96 | .38 |
| Perceived MC | | | | 1487.11 | 884.32 | .26 | 535.90 | 848.40 | .16 |
| Product/Process MC | | | | | | | 1003.68 | 345.14 | .76* |
| R² | .33 | | | .40 | | | .54 | | |
| Adjusted R² | .31 | | | .35 | | | .49 | | |
| F | 13.94** | | | 8.84** | | | 10.34** | | |
| R² Δ | .33** | | | .07 | | | .15* | | |

Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively.

Table 4.4. Factors Predicting Body Weight Status

| | Model 1 | | | Model 2 | | | Model 3 | | | Model 4 | | |
|-------------------------------|---------|------|---------|---------|------|---------|---------|------|---------|---------|------|---------|
| | B | SE | β | B | SE | β | B | SE | β | B | SE | β |
| (Constant) | 6.54 | 4.34 | | 5.61 | 5.45 | | -5.69 | 7.89 | | 1.94 | 9.50 | |
| Years of Age | 1.20 | .34 | .55* | 1.20 | .35 | .55* | 1.73 | .43 | .79** | 1.65 | .67 | .76** |
| Perceived Motor competence | | | | .33 | 1.15 | .05 | 1.50 | 1.25 | .21 | 1.58 | 1.23 | .22 |
| Product/Process MC | | | | | | | -1.05 | .55 | -.42 | -.51 | .67 | -.20 |
| Physical Activity | | | | | | | | | | 0.00 | .00 | -.29 |
| R² | .30 | | | .31 | | | .39 | | | .44 | | |
| Adjusted R² | .28 | | | .26 | | | .32 | | | .34 | | |
| F | 12.23* | | | 5.96* | | | 5.57* | | | 4.81* | | |
| R² Δ | .30* | | | .00 | | | .09* | | | .04 | | |

8 Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively.

included physical activity. Model 1, level 1 significantly accounted for 27% of the variance ($R^2 = .27, p = .013$) in HRQoL with the participants demographics, co-morbidity ($\beta = -.37, p = .033$; 1 = co-morbidity and 0 = no co-morbidity) and biological sex ($\beta = -.32, p = .066$; 1 = female). Model 2 did not significantly predict the participants HRQoL see Table 4.13. Model 3, level 1 co-morbidity ($\beta = -.37, p = .019$) and biological sex ($\beta = -.26, p = .092$), level 2 perceived motor competence ($\beta = .25, p = .11$), and level 3, motor competence process/product ($\beta = .40, p = .009$) predicting the participants total HRQoL scores. A total of 51% of variance in participants' total HRQoL scores was explained in model 3 with motor competence adding an additional 16% of variance explained ($R^2 \Delta = .16, p = .009$) beyond co-morbidity, biological sex, and perceived motor competence. In model four, physical activity ($\beta = .16, p = .38$) did not add a significant amount of variance explained ($R^2 \Delta = .02, p = .375$). See Table 4.6. for the complete results of the four-level hierarchical regression. A post-hoc priori power analysis was conducted using G*Power Software version 3.1.9.4 for a four-level hierarchical regression by imputing an alpha level of 0.05, observed $R^2 = .52$, participants ($N = 30$), three measured predictors, with five total predictors (1- β err prob = .99).

Discussion

The purpose of this study was to examine the underlying mechanisms predicting physical activity and HRQoL above and beyond significantly correlated demographics (e.g., etiology), for youth with a VI or VISC. The physical activity levels for this sample were only significantly predicted by degree of vision and motor competence, not by perceived motor competence, although model three as a whole was significant. Overall, the hypothesis of the physical domain predicting HRQoL was not completely supported,

Table 4.6. Factors Predicting Health-Related Quality of Life

| | Model 1 | | | Model 2 | | | Model 3 | | | Model 4 | | |
|-------------------------------|---------|------|---------|---------|-------|---------|---------|-------|---------|---------|-------|---------|
| | B | SE | β | B | SE | β | B | SE | β | B | SE | β |
| (Constant) | 87.77 | 3.45 | | 70.83 | 10.44 | | 75.27 | 11.83 | | 68.81 | 11.83 | |
| Co-morbidity | -11.64 | 5.20 | -.37 | -10.06 | 5.11 | -.32 | -11.45 | 4.55 | -.37* | -11.29 | 4.57 | -.36* |
| Biological Sex | -9.58 | 5.00 | -.32 | -7.43 | 4.99 | -.25 | -7.76 | 4.24 | -.26 | -7.75 | 4.45 | -.36 |
| Perceived Motor competence | | | | 5.35 | 3.13 | .29 | 4.68 | 2.78 | .25 | 3.79 | 2.96 | .20 |
| Product/Process MC | | | | | | | 3.94 | 1.39 | .40* | 3.25 | 1.60 | .33 |
| Physical Activity | | | | | | | | | | 0.00 | .001 | .16 |
| R² | .27 | | | .35 | | | .51 | | | .52 | | |
| Adjusted R² | .22 | | | .27 | | | .43 | | | .42 | | |
| F | 5.07* | | | 4.60* | | | 6.40** | | | 5.25* | | |
| R² Δ | .27* | | | .07 | | | .16* | | | .02 | | |

Note: * and ** indicate a significant difference between each group, $p < .05$ and $p < .001$ respectively.

however motor competence was a strong and significant predictor of HRQoL. Therefore, further research needs to be conducted to suggest that the Stodden and colleagues model (2008) may predict more than just physical activity and BMI for youth with a VI or VISC.

Correlations were examined for both participant demographics and assessed variables of the physical domain and HRQoL, in which some were supported by literature and others contrasted with previous findings. Participant characteristics that were significantly associated and supported in the literature included: the positive relationship between age and BMI (CDCP, 2020; De et al., 2008), degree of vision with motor competence (Haibach et al., 2014) and degree of vision with physical activity (Brian et al., 2019a). An expected finding, due to a secondary data analysis, was the lack of any relationship with the participants etiology (e.g., VI or VISC; supported by the findings in study 2 (Gilbert et al., In Preparation). Although, in the general population a positive relationship with age and motor competence has been conceptualized and empirically supported (Cairney et al., 2019; Robinson et al., 2015; Stodden et al., 2008), this relationship between age and motor competence has not been supported historically in the literature on youth with VIs. Meaning that youth with VIs are not improving their motor competence abilities over time, while youth in the general population are improving in motor competence across time. While a significant negative relationship between co-morbidity and HRQoL was expected, it was unexpected that it was not significantly associated with other variables such as motor competence. Children with co-morbidities have been historically overlooked or not identified in the literature regarding the physical domain for youth with a VI, however one cannot assume a child with only a VI will perform the same as a child with a VI and an intellectual disability or another condition, supported

by our findings and should continue to be further explored. The only other participant demographic that was significantly associated with overall HRQoL was biological sex, showing lower scores for HRQoL were more likely to be female participants and higher scores were more likely to be male participants. This is a major concern that our female youth are demonstrating a significantly lower HRQoL than that of their male peers also with a VI. The biological sex differences in other self-perceptions are supported in literature in the VI population (McMahon et al., 2019), but findings on biological sex differences in adults with VI on quality of life were not significant (Holbrook et al., 2009). This finding of no biological sex differences in adults with a VI on quality of life contrasts the relationship results of this study between HRQoL and biological sex, and may suggest that this relationship changes over time, and should be further examined with longitudinal studies.

The relationships with our predicted variables, which were used to guide the hierarchical regressions, were mostly aligned with the current literature. Participants' physical activity had a strong to moderate relationship with degree of vision, motor competence, and HRQoL, all of which are supported in the general and/or VI populations (Brian et al., 2019a, 2019b; Wu et al., 2017). Unexpectedly, our results included perceived motor competence not being significantly associated with physical activity, contrasting the relationships in the literature on the general population (Babic et al., 2014; Brian et al., 2019b; True et al., 2017). Total HRQoL scores were significantly associated with biological sex, co-morbidity, physical activity, perceived motor competence, and motor competence (product and process). Relationships with HRQoL that are supported in the literature include co-morbidity (Boulton et al., 2006), and biological sex (McMahon et al.,

2019). However, Holbrook and colleagues (2009), found HRQoL could not be predicted in adults with VI by physical activity levels or degree of vision. To our knowledge no one has looked at the relationship or predictive strength of perceived motor competence to HRQoL in youth with VI or VISC. Motor competence product consistently had stronger relationships with the variables of interest than the TGMD-3; which aligns with research suggesting that product may be a better predictor of perceived motor competence than process motor competence assessments (True et al., 2017, 2019). The relationship between product motor competence and physical activity ($r = .71$) was to our knowledge the highest indicated in the literature, Brian and colleagues (2019a) found motor competence to be moderately associated ($r = .41$ object control and $r = .40$ locomotor) with physical activity for youth with VIs. To increase confidence in the strength of product motor competence measures over process measures, the study needs to be replicated and scaled up to a larger participant size, and utilizing different process-oriented tests.

To answer the first research question of this study, the demographic variables (degree of vision) that were significantly associated with physical activity were imputed in level 1 of the regression. Degree of vision significantly predicted physical activity, which aligns with the literature on youth with VIs (Brian et al., 2019a). Motor competence (process and product) was also a significant predictor in overall physical activity which appears to align with the 2008 conceptual model of the relationship between motor competence and physical activity (Robinson et al., 2015; Stodden et al., 2008). However, perceived motor competence was not a significant predictor in physical activity, contrasting the conceptual model and the literature in the general population and youth with disabilities that suggests perceived motor competence will act as a mediator

(Robinson et al., 2015; Stodden et al., 2008) or may be the strongest predictor (Babic et al., 2014; Brian et al., 2019b). The predictive strength of perceived motor competence on physical activity levels in youth with a VI or VISC should be further explored with physical activity measured in a more normal setting than a sports camp, such as a regular school week at home.

Targeting the second research question of this study, the demographic variables (biological sex and co-morbidity) that were significantly associated with HRQoL were imputed in level 1 of the regression. Participants' presence or absence of a co-morbidity and their biological sex significantly predicted HRQoL. Above and beyond the participants' demographics, motor competence significantly predicted HRQoL, aligning with our hypothesis. However, perceived motor competence and physical activity did not significantly add to the model even though the associations were moderate. This finding may be due to insufficient power from a small sample size and should be further examined. Overall, the findings indicate promise to the 2008 Stodden and colleagues conceptual model predicting more than physical activity and body weight status.

There were limitations to this study, of which the reader should be aware of when interpreting the findings. Due to the nature of low incidence of a VI or VISC, only a small convenience sample could be obtained. Other studies on youth with VIs or VISC examining the physical domain have also had small samples (14 to 46 participants), due to the low incidence population (Colgan et al., 2016; Haegele et al., 2017; Lieberman & McHugh, 2001). Therefore, if possible a larger sample size should be acquired for more power in the future. Additionally, the participants' physical activity levels may have been strongly influenced by the testing site (school or a sports camp). This could have impacted

the predictive power of physical activity, as well as its relationship with other variables and participant characteristics. However, Brian and colleagues (2019a) reported a moderate association ($r = .35$) with participants physical activity recall and physical activity at a sports camp for the blind and VI. The setting of an educational sports camp may also impact how representative the sample was to youth with a VI or VISC populations. Additionally, even though the presence of a co-morbidity was added to the model for predicting HRQoL it is unknown if the co-morbidity was a direct link to the cancer or treatment. Finally, self-report surveys were used to collect the data for perceived motor competence and HRQoL, which may have been influenced by a camp atmosphere and contribute to biased responses. This study should be replicated with participants outside of a camp setting in the future.

There are many implications for practitioners based on the findings of this study. First, given that motor competence was the strongest predictor of both physical activity and HRQoL in youth with VI and VISC, physical educators might need to emphasize motor skill development if the long-term goals are to support physical activity levels and overall HRQoL for youth with VI and VISC. Second, practitioners are encouraged to be aware of the potential impact individual characteristics (e.g., biological sex, co-morbidity, and degree of vision) may have on their students' levels of physical activity and HRQoL. Specifically, children with lower levels of vision are more likely to be less physically active and females or youth with a co-morbidity are more likely to have lower levels of overall HRQoL. These students must be fully included in the same curriculum as their same age peers in order to gain the same benefits for a lifetime of sport and physical activity and ensure the best HRQoL possible.

In conclusion, the physical domain has a strong relationship with overall HRQoL for youth with a VI or VISC. Above and beyond participant characteristics, motor competence was the strongest predictor for both physical activity and HRQoL. Therefore, practitioners and researchers should be aware of the importance not only of physical activity, but specifically of motor competence and its impact on HRQoL for youth with VIs and VISCs. The findings in this study indicate an importance of future research that looks further into the physical domain and the potential impact it may have on long-term HRQoL.

CHAPTER 5:

DISCUSSION

This dissertation was comprised of three studies targeting the motor competence and HRQoL for youth with pediatric cancers, a VI, and VISC. The overall purpose was to examine the motor competence of youth with pediatric cancers, a VI, and VISC and explore if motor competence and other variables in the physical domain predict overall HRQoL. Specifically, Study 1 was a systematic review of literature exploring the gross motor competence of pediatric cancer patients and survivors in comparison to healthy peers, and the correlates of motor competence for this population (e.g., HRQoL, physical activity, perceived motor competence). Study 2 examined the differential effects of healthy peers, individuals with a VI or VISC on HRQoL, motor competence (process and product), perceived motor competence, and physical activity levels. Finally, Study 3 examined the underlying mechanisms predicting physical activity and HRQoL for youth with a VI or VISC, beyond significantly correlated participant characteristics (e.g., degree of vision).

Each of these studies addressed substantial gaps in the literature regarding motor competence, perceived motor competence, physical activity, and HRQoL for youth with a pediatric cancer, VI, or VISC. Study 1 served as an update and expansion on our current understanding of motor competence of pediatric cancer survivors and patients, and correlates of their motor competence. Study 2 addressed two major gaps in the literature: 1) what is the motor competence, perceived motor competence, physical activity level, and

HRQoL for youth with a VISC, and 2) how do youth with a VISC differ from their healthy or visually impaired peers. Study 3 addressed the knowledge gaps regarding the associations among relationships between motor competence, perceived motor competence, physical activity and HRQoL for youth who have or had a VI, or VISC in comparison to their peers without either. Additionally, Study 3 explored how these variables (motor competence, perceived motor competence, and physical activity) predict physical activity and overall HRQoL in these populations of youth with a VI and VISC. The findings of these studies will help to bridge gaps in the knowledge base regarding motor competence, perceived motor competence, physical activity, and HRQoL for youth with pediatric cancer, a VI, or VISC.

This dissertation is filling a significant need, due to its potential to predict and improve HRQoL for youth with a VI and VISC through the physical domain (e.g., motor competence). The methodologies used throughout these studies are innovative as they are the first to examine the motor competence, physical activity, perceived motor competence, and HRQoL of individuals with VI and VISC. Guiding this study was a conceptual model of the relationship between motor competence and physical activity (Stodden et al., 2008). Another innovative aspect of this study is its examination of this model and its prediction of not only body weight status, but HRQoL. This dissertation was conducted using rigorous methodology that included a combination of process and product measurements of motor competence as recommended by experts in the field.

Each of the studies was designed to sequentially and methodologically build upon one another, while the research questions being targeted were developed based on the findings of the previous study. Study 1 was a systematic review of the literature on motor

competence and correlates of motor competence for pediatric cancer survivors. Based on the findings from the systematic review in Study 1, youth previously excluded from pediatric cancer literature with a VISC became the focus of Study 2. Additionally, HRQoL was used as the major outcome with limited information particularly in the physical domain in these populations for Study 2. After analyzing the findings in Study 2, participants with a VI and VISC were not significantly different from one another on any of our variables of interest and therefore were not controlled for in Study 3. Overall, the sequential order of these studies follows sound research practices for answering questions within a line of inquiry.

After qualitatively analyzing the studies included in the systematic review, following PRISMA guidelines, in Study 1, the empirical evidence indicates that overall the motor competence in pediatric cancer survivors is significantly lower than that of same-aged healthy peers. This finding aligns with a review previously completed on motor competence that only included children with acute lymphoblastic leukemia (Green et al., 2013). Additionally, physical activity, BMI, grip strength, and age were significantly correlated with motor competence for pediatric cancer survivors (Götte et al., 2015; Ness et al., 2015; Piscione et al., 2017; Sabel et al., 2016; Wright et al., 2005). Irrespective of time since the completion of treatment, (e.g., three to ten years) most studies found that pediatric cancer survivors' post-treatment are still exhibiting motor delays, indicating a need for intervention (De Luca et al., 2013, Hartman et al., 2006; Kesting et al., 2015; Leone et al., 2014; Piscione et al., 2014; van Brussel et al., 2006). Thus, physical educators can potentially provide a key role in the future developmental trajectory of health for this vulnerable population.

Within Study 2, exploring group differences multiple interesting findings appeared, although the overall hypothesis was not supported. However, both the VI and VISC groups were consistently below their comparison groups in motor competence, but not significantly different from one another. Contradicting previous literature, the comparison groups mean HRQoL was much lower than both the VI and VISC groups (Boulton et al., 2006; Chadha & Subramanian, 2011; Chen et al., 2005; Gohar et al., 2011; Habib & Irshad, 2018; Ness et al., 2015; Wong et al., 2009; Wright et al., 2005). Additionally, the average physical activity for the comparison group was significantly lower than that of the VI and VISC group. This opposing finding may have been due to the VI and VISC groups wearing their Fitbits™ during a week-long summer sports camp, and the comparison group wearing their Fitbits™ during a typical school week (Haegele & Porretta, 2015). In summary, the motor competence, perceived motor competence, physical activity, HRQoL of youth with VIs and a VISC are not significantly different from one another, but are lower than peers without a VI or VISC.

The findings in Study 3 indicated motor competence as a significant predictor in both physical activity and HRQoL. Although, perceived motor competence was not a significant predictor in either physical activity or HRQoL contrasting historical findings (Babic et al., 2014; Brian et al., 2019b), it was moderately correlated with HRQoL and may have lacked power for sensitivity to predict. Participant characteristics that were significantly associated with HRQoL included biological sex and co-morbidity which are supported in literature in the general and VI populations (Boulton et al., 2006; McMahon et al., 2019). Overall, the hypothesis of the physical domain predicting physical activity and HRQoL was supported. This appears to align with the 2008 conceptual model

regarding the relationship between motor competence and physical activity that suggests the model may predict more than just physical activity for youth with VI or VISC (Robinson et al., 2015; Stodden et al., 2008).

The major findings of this dissertation include the strength of motor competence in predicting both physical activity and HRQoL for youth with a VI or VISC. Additionally, physical activity and perceived motor competence had significant moderate relationships, which may predict HRQoL with a larger sample size. Another major finding was that youth with a VI and VISC were not significantly different from one another, but were from their same-aged peers without a VI. This study also examined motor competence with a combination of process- and product- oriented measurements that have previously been recommended in the general population (Burton & Miller, 1998; Hands, 2002; True et al., 2017). As dictated by the results of this study, product-oriented measurements of motor competence showed higher levels of correlation and predictive strength for both HRQoL and physical activity levels than the TGMD-3 showed.

Implications for Practitioners

Based on the results of this dissertation, the importance of motor competence should be emphasized in children's overall levels of physical activity and perceived HRQoL, which can have a large impact on health outcomes. Thus, physical education focused not only on physical activity and fitness, but on motor competence can potentially provide a fundamental role in the future developmental trajectory of health for this vulnerable population. Practitioners should note that based on these results the most significant individual characteristics for predicting physical activity and HRQoL were degree of vision, biological sex, and co-morbidity. When planning and instructing students

with a VI and VISC, content should be focused on motor competence development and individual characteristics should be recognized.

Implication for Future Research

The implications of this dissertation open a line of inquiry that can lead to the long-term goal of improving HRQoL for individuals with a VI and VISC. Although, group differences between individuals with a VI and VISC were not significant, differences between ocular and cortical VIs should be explored in the future. Though not significant, the VISC groups mean scores on both product (locomotor and ball skills) and process (locomotor and ball skills) motor competence were higher than that of those in the VI group. Being that a VISC is an acquired VI, it was noted that most of the participants' VIs in the VI group were congenital. Therefore, future research should also examine the participants' age of onset of VI, which may be a stronger indicator of motor competence than the etiology of the VI.

Based on the promising findings of this dissertation, future research should continue to examine the impact of motor competence and the physical domain in overall HRQoL for youth with a VI and VISC. The development of motor competence targeted interventions should be tailored for these populations, based on the individual characteristics of biological sex, co-morbidity, and degree of vision. Additionally, we recommend that future studies regarding the physical domain for youth with a VI or VISC do not ignore the presence of a co-morbidity. The use of the Stodden and colleagues (2008) model should also be confirmed within this population and examine if these relationships and predictions within the model hold over time for physical activity and HRQoL.

Conclusion

The overall results indicated that youth with a VI and VISC, although not significantly different from one another, differ from their sighted peers in motor competence and perceived motor competence. The relationships conceptualized in the Stodden and colleagues (2008) model are promising in this population and should be examined further with a larger sample. Additionally, practitioners and researchers should be aware of individual characteristics that may impact the motor competence, physical activity, and HRQoL such as, degree of vision, biological sex, and co-morbidity for youth with a VI and VISC. Moving forward the presence of a co-morbidity in youth with a VI or VISC should be controlled for in future research. In conclusion, the findings of this dissertation suggest and support the importance motor competence plays in youth with a VI or VISC on HRQoL and implore that this interaction no longer be overlooked.

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